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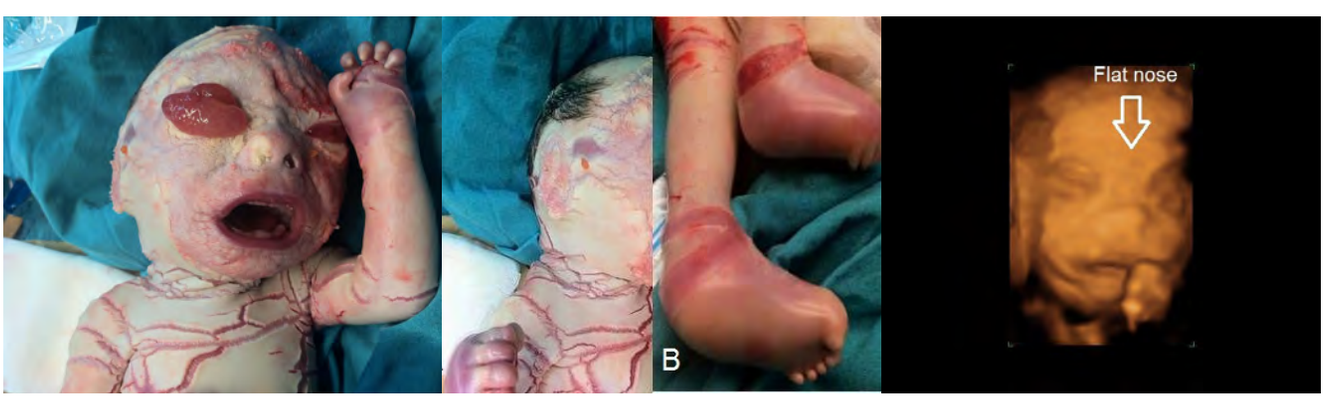
Prenatal Diagnosis of Harlequin Ichthyosis: Report of a Case

Sanhal CY, Özekinci M, Sakıncı M, Şimşek M, Mendilcioğlu İ



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JOURNAL OF CLINICAL AND ANALYTICAL MEDICINE

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Cor Triatriatum with Ankylosing Spondylitis

Ankilozan Spondilit ile Birlikte Cor Triatriatum

M. Onur Hanedan, A. İhsan Parlar, Seyhan Babaroğlu, Kerem Yay, Garip Altıntaş
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Özet

Cor triatriatum özellikle sol atrium olmak üzere atrial boşluklardan birinin fibromusküler membran ile ikiye bölünmesiyle oluşan nadir bir konjenital defekttir. Ankilozan spondilit kardiyak tutulumunda görülebildiği kronik sistemik inflamatuvar romatizmal hastalıktır. Ankilozan spondilit ile takip edilen 27 yaşında kadın, kardiyak tutulum için taranırken, cor triatriatum ekokardiyografi ile teşhis edildi ve tanı MRI ve CT anjio ile doğrulandı. Membran kardiyoplejik arrest ile kardiyopulmoner baypas altında cerrahi olarak çıkarıldı.Hasta şifa ile hastaneden taburcu edildi. Sonuç olarak, Ankilozan spondilit'li hasta kardiyak tutulum için taranmalıdır.

Anahtar Kelimeler

Cor Triatriatum; Ankilozan Spondilit

Abstract

Cor triatriatum is a rare congenital defect that one of the atrial chambers especially left atrium, is divided into two by a fibromuscular membranee. Ankylosing spondylitis is a chronic systemic inflammatory rheumatic disorder that cardiovascular involvement can be seen. 27-year-old woman, who has been followed for ankylosing spondylitis searched for cardiac involvement, Cor triatriatum was diagnosed with echocardiography and diagnosis was confirmed with MRI and CT angiography. Membrane was resected surgically under cardiopulmonary bypass with cardioplegic arrest. Patient was discharged from the hospital with recovery. In conclusion, patient with ankylosing spondylitis must be searched for cardiac involvement.

Keywords

Cor Triatriatum; Ankylosing Spondylitis

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Introduction

Cor triatriatum is a rare congenital defect that one of the atrial chambers is divided into two by a fibromuscular membrane. It is found 0.4% in a series of 474 patients' autopsy with congenital heart defect (1). More often patients present in infancy but some cases can remain asymptomatic until adulthood. Morphology of cor triatriatum left atrium division is more common. Cor triatriatum sinister is only 0.1% of the congenital heart diseases (2).

Ankylosing spondylitis is a chronic systemic inflammatory rheumatic disorder that mainly involves young males and primarily effects on axial skeleton. Peripheral joints, tendons and ligaments may be affected. Uveitis, cardiovascular and pulmonary involvement may also occurs (3). Cardiovascular involvement is found 42.5% in a retrospective study (4). Aortic insufficiency, atrio-ventricular block, bundle branch block, Wolff-Parkinson-White syndrome, short PR syndrome, ischemic heart disease, mitral regurgitation, diastolic dysfunction (left ventricular dysfunction), tachy-brady syndrome, atrial fibrillation are cardiac involvement (4, 5). However, coexisting with cor triatriatum did not reported previously.

Case Report

27-year-old woman, who has been followed for ankylosing spondylitis from March 2006 with pelvic articulation involvement, has no cardiac symptoms. On examination, the patient was found to be normotensive with a regular pulse and a normal electrocardiogram. A transthoracic echocardiography was performed to search for, if there is any cardiac involvement of ankylosing spondylitis. It is showed that there was a membrane below 1.3 cm of the mitral valve and had an 11/5 mmHg gradient, 2 degree mitral insufficiency with a 45 mmHg systolic pulmonary artery pressure. After that, a transoesophageal echocardiography was performed which showed a membrane in the left atrium between left atrial appendix and left upper pulmonary vein (Figure 1). Cardiac magnetic resonance imaging and cardiac computed tomography angiography were performed, both were reported as cor triatriatum sinister (Figure 2a and 2b).

Surgical Technique

In the general anesthesia, median sternotomy was performed. After the heparin was administered, total cardiopulmonary bypass (CPB) was established between the ascending aorta and both superior vena cava and inferior vena cava. Under moderate hypothermic conditions, CPB flow rate was maintained at 2.5 L/min/m², aorta was cross-clamped and cardiac arrest was maintained with antegrade cardioplegia. Left atriotomy was performed and a membrane, which had 0.5 cm2 opening, was noticed in the left atrium. Membrane was resected. Mitral valve was competent. There was no atrial septal defect. Left atriotomy was closed. Intraoperative transesophageal echocardiography was performed which shown no abnormalities after resection . Postoperative period was uneventful and patient was discharged from the hospital with healthy.

Discussion

Cor triatriatum is a rare congenital defect. Morphologically left atrium division is more common. More often patients present in infancy, but some cases can remain asymptomatic until adulthood (1,2). Ankylosing spondylitis is a chronic systemic inflammatory disorder that cardiovascular involvement can be seen (3). This involvement was reported about 42.5% in a retrospective study (4). This involvement generally makes valvular insufficiency and rhythm disorders due to involvement of conduction system in the heart. This is the first case of cor triatriatum sinister with ankylosing spondylitis in the literature.

It is important to search ankylosing spondylitis patients for cardiac involvement. Electrocardiography and echocardiography must be performed to search for any arrhythmias', possible valve abnormalities, and congenital disorders (5).

The treatment is surgery when cor triatriatum is found. Surgical treatment of cor triatriatum is a membrane resection and repair of concomitant abnormalities.

In conclusion, patient with ankylosing spondylitis must be searched for cardiac involvement.

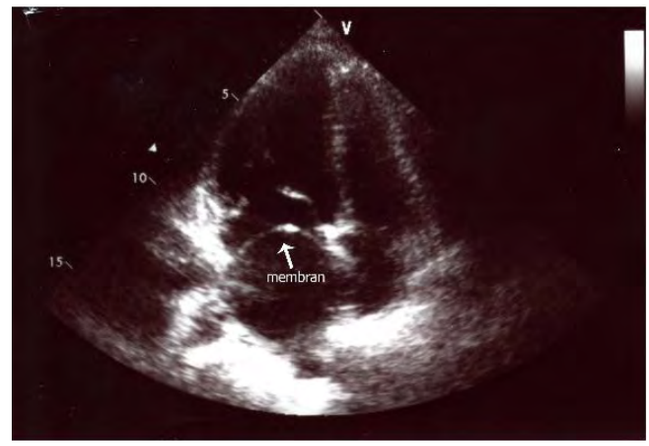

Figure 1. View of the preoperatif transthoracic echocardiography, arrow indicate membrane in the left atrium.

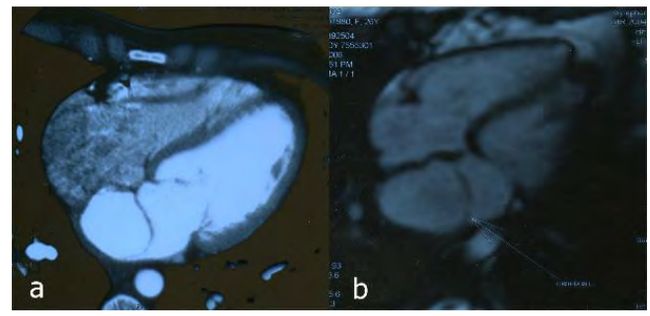

Figure 2. View of the membrane with Magnetic Resonance Imaging (A) and Computed Tomography Angiography (B).

Competing interests

The authors declare that they have no competing interests.

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Case Report
JCAM

Ischemic Changes in a Case of Unilateral Pseudoexfoliation Syndrome

Tek Taraflı Psödoeksfoliasyon Sendromlu bir Olguda İskemik Değişiklikler

Ischemic Changes in Pseudoexfoliation Syndrome

Tolga Kocaturk[1], Isıl Isıklıgil[1], Volkan Dayanir[1], Yelda Ozsunar[2], Harun Cakmak[1]
[1]Department of Ophthalmology, [2]Department of Radiology, Adnan Menderes University Medical School, Aydin, Turkey

Özet

66 yaşında, glokomatöz görme alanı kayıplarını taklit eden, normotansif, tek taraflı psödoeksfoliasyon sendromlu bir erkek hasta sunulmaktadır. Hastada, tek taraflı belirgin iskeminin eşlik ettiği retinal sinir lifi tabakasında incelme ve ardışık görme alanlarında nazal adım defektleri vardı. Klinik olarak psödoeksfoliasyonun görüldüğü sol gözde, optik disk görünümü glokomatöz çukurlaşmayı desteklemese de; sağ göze göre daha soluk ve retinal damarlar daralmış görünümdeydi. Sol oftalmik arterin renkli Doppler görüntülemesinde, rezistivite indeksinin 0.88 gibi oldukça yüksek bir değerde olduğu görüldü. Olgu, psödoeksfoliasyonun oküler iskeminin bir sebebi olduğunu belirten yeni literatür bilgileri ışığında tartışıldı.

Anahtar Kelimeler

Psödoeksfoliasyon Sendromu; Renkli Doppler; Retina; Sinir Lifi Tabakası; İskemi

Abstract

A 66 year old man with normotensive unilateral pseudoexfoliation syndrome associated with ipsilateral marked ischemia with nerve fiber layer thinning and nasal step on successive visual field tests mimicking glaucomatous visual field loss is presented. Although the optic disc appearance of the clinically visible pseudoexfoliative left eye was not suggestive of glaucomatous cupping the disc appeared much pale and retinal vessels narrowed compared to the right eye. Color Doppler imaging of the left ophthalmic artery showed extremely high resistivity index of 0.88. The case is discussed in light of recent literature underscoring the fact that pseudoexfoliation is a cause of ocular ischemia.

Keywords

Pseudoexfoliation Syndrome; Color Doppler; Retina; Nerve Fiber Layer; Ischemia

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Introduction

Pseudoexfoliation syndrome (PXS) is the most common identifiable cause of open angle glaucoma worldwide [1]. Pseudoexfoliative material on the walls of iris vessels, posterior ciliary arteries, vortex veins, and central retinal vessels [2] may alter blood flow parameters by causing several types of vascular damage [2,3].

Accumulation of pseudoexfoliative material in iridial vessel walls is associated with increased permeability, narrowing, and finally obstruction [2]. Increased ophthalmic artery resistivity index (RI) in pseudoxfoliation have been reported by two independent studies [4,5]. Saatçi et al. found presence of pseudoexfoliative material as a likely risk factor for retinal vein occlusion [6]. Puska et al. observed optic disc changes in the affected eyes of unilateral PXS patients over time and concludes that the pseudoexfoliative process itself may be a risk factor for optic disc changes [7].

We here in describe a normotensive case of unilateral PXS with marked increase in RI of the ipsilateral ophthalmic artery and significant retinal vascular attenuation associated with optic disc pallor and a visual field defect resembling nasal step in the left eye. To the best of our knowledge, this is the first case of optic neuropathy associated with marked ischemia mimicking glaucomatous visual field loss in a patient having PXS.

Case Report

A 66 year old man presented to our outpatient clinic with a chief complaint of itching, burning and stinging in both eyes. He had no prior medical history of diabetes mellitus, hypertension or family history of glaucoma. He was not using any chronic topical or oral medication. Best corrected visual acuities were 0.00 logMAR equivalent with +1.00 D refraction in the right, and 0.10 logMAR equivalent with +1.00 (+0.25x110) refraction in the left eye. Intraocular pressures (IOP) measured with a non contact tonometer (Reichert, Xpert NCT Plus, Buffalo, NY) on the day of outpatient visit were 13 mmHg and 12 mmHg on the right and left eyes respectively. Biomicroscopic examination showed pseudoexfoliative material at the pupil margin of the left eye. Dilated fundus examination showed cup-to-disc ratio to be 0.35 and 0.3 in the right and left eyes respectively, there were no nerve fiber layer defect or disc notching, and vertical disc size was 1.1 mm (as measured using a +78D lens) in both eyes.

The patient was considered to be unilateral PXS and was planned to be recruited in a clinical study. Office-hour IOPs were 15, 13, 12 mmHg, and 14, 13, 12 mmHg for the right, and left eyes respectively with Goldmann applanation tonometry. Corneal pachymetry (IOPac Advanced Pachymeter, Heidelberg Engineering, StarFish, Victoria, BC, Canada, V8V 2T2) in the right, and left eyes were 541 ± 3.5 µm, and 554 ± 2.0 µm respectively. The visual field test that was repeated three times within 4 months was normal for the right eye whereas the left eye consistently had a superior nasal step. The last follow-up visual field had been done in 17th month follow-up (Figure 1).

Due to the superior nasal step visual field defect observed in the left eye, the diagnosis of "unilateral PXS" was reconsidered and the patient had a second dilated fundus examination. Significant disc pallor especially prominent inferotemporally corresponding to the superior nasal step visual field defect as well

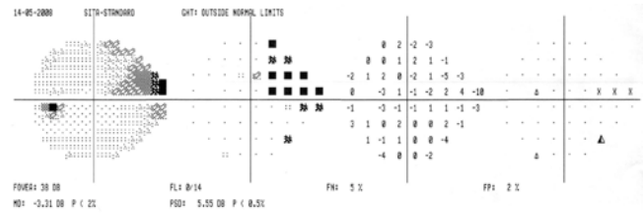

Figure 1. Left eye had a superior nasal step that was consistent in all visual field tests and Glaucoma Progression Analysis of the last visual field test showing no progression

as decreased caliber and irregular contour of all retinal vessels were observed in the left eye compared to the right (Figure 2).

Scanning laser polarimetry (GDx VCC, Carl Zeiss, USA) (Figure 3) and optical coherence tomography (OCT/SLO, Ophthalmic

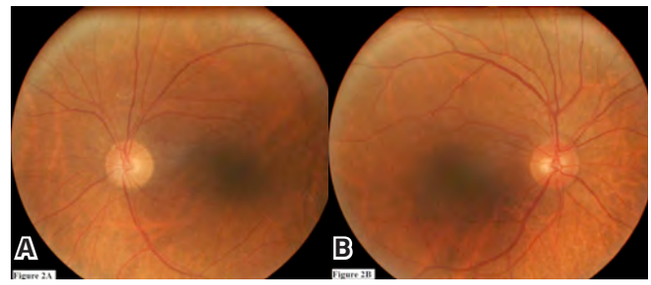

Figure 2. (A) Left fundus showing a c/d 0,3, pale optic disc and vessels of decreased caliber compared to the right eye. (B) Right fundus showing a c/d 0,35.

Technologies Inc., Canada) (Figure 4) revealed thinned nerve fiber layer in the left eye.

The subject had normal color vision with Ishihara plates and no relative afferent pupillary defect.

Brain and orbital magnetic resonance imaging (MRI) revealed non-specific periventricular gliosis and mild thinning of the left

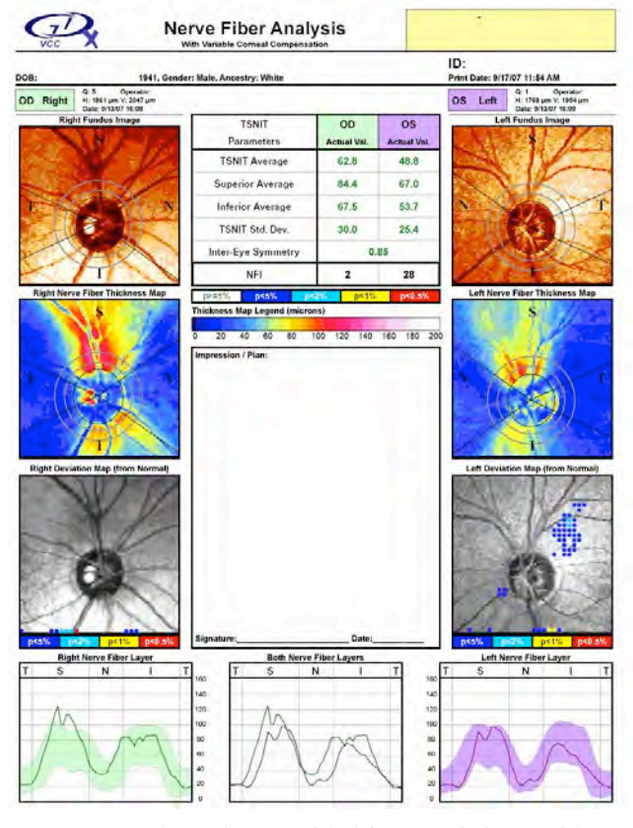

Figure 3. Scanning laser polarimetry of the left eye reveals thinning of the nerve fiber layer inferiorly although it is within normal limits.

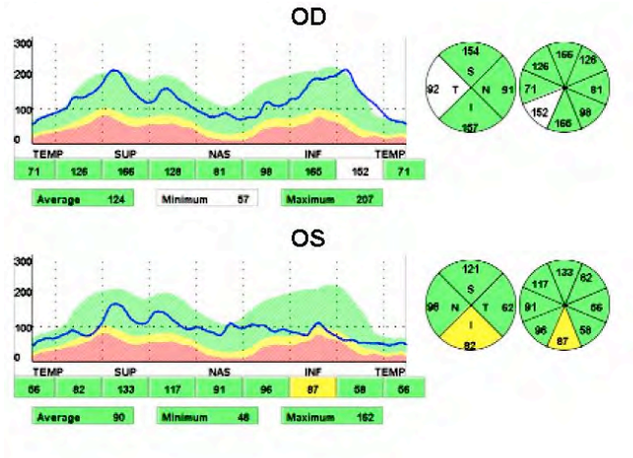

Figure 4. Optical coherence tomography showed nerve fiber layer thinning in the left eye.

optic nerve.

Color Doppler imaging of ophthalmic artery showed peak systolic velocity (PSV) of 24.9 cm/s and 42.1 cm/s, end diastolic velocity (EDV) of 7.2 cm/s and 5.0 cm/s, and RIs of 0.71 and 0.88 in the right and left eyes respectively.

Discussion

The RI is a comparison of the flow in systole and diastole and provides an indication of the resistance in the peripheral vascular bed; it can be used to facilitate an evaluation of perfusion of an organ [8]. We have previously shown that in unilateral PXS, the affected side has significantly higher mean ophthalmic artery RI (mean ± SD, 95% CI; 0.74 ± 0.04 cm/s, 0.72-0.75) compared to age-sex matched controls (0.69 ± 0.07 cm/s, 0.66-0.72; p=0.009) [5]. Similar results were obtained by Yüksel et al. [4] We have suggested a value of 0.72 for RI to be the cut-off value for differentiation of PXS.

The case in discussion had a left ophthalmic artery RI of 0.88 which is far above what we had reported for the unilateral PXS [5]. We believe that the attenuated retinal vessels and pale optic disc seen on the left fundus are a reflection of increased vascular resistance in the ophthalmic artery due to buildup of pseudoexfoliative material in vessel walls. The chronic ischemia might have led to progressive thinning of the nerve fiber layer which might eventually lead to marked cupping of the optic disc. Indeed, presence of thinner retinal nerve fiber layer has been shown in PXS when compared to the controls [9].

Repo et al. [10] suggested pathologic changes in the blood supply of PXS eyes by showing strong correlations between PSX and patients who had had a transient ischemic attact with abnormal iris transluminance.

We believe that the case described is an extreme example of normotensive ocular ischemia caused by pseudoexfoliation. Such cases are probably missed on clinical grounds since they are normotensive and are not given routine visual fields or other ancillary glaucoma tests. We suggest routine glaucoma work-up to all pseudoexfoliation patients regardless of their IOPs and use the ophthalmic artery RI of 0.72 as the cut-off value when evaluating increased vascular resistance for ophthalmic artery.

Competing interests

The authors declare that they have no competing interests.

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Pilomatrixoma of the Lower Extremity

Alt Ekstremitede Pilomatriksoma

Tibial Pilomatrixoma

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Özet

Trikomatriksoma, Trikilemmal kist veya Malherbe'nin kalsifik epitelyoması olarakta bilinen Pilomatriksoma kıl foliküllerinden köken alan iyi huylu bir tümördür. Çok nadir görülmesine karşın yanlış tanınabilmesi ve diğer lezyonlarla karışabilmesi nedeniyle klinik öneme sahiptir. Pilomatriksoma genelde boyun bölgesinde görülmektedir. Bu olgu sunumunda daha once literatürde çok nadir görülen alt ekstremite yerleşimli pilomatriksoma anlatılmaktadır.

Anahtar Kelimeler

Pilomatriksoma; Alt Ekstremite; Benign Tümör

Abstract

Pilomatrixoma, also known as trikomatriksoma, trichilemmal cyst, or Malherbe's calcifying epithelioma, is a benign tumor originating from the matrix cells of the hair follicles. Despite its rarity, it is of clinical importance, as it may lead to incorrect pre-diagnosis, being confused with other lesions. It is generally seen on the neck region. In this case report, pilomatrixoma of the lower extremity has been described, which has been described in the literature extremely rare.

Keywords

Pilomatrixoma; Lower Extremity; Benign Tumor

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Introduction

Pilomatrixoma, also known as trikomatriksoma, trichilemmal cyst, or Malherbe's calcifying epithelioma, is a benign tumor originating from the matrix cells of the hair follicles. Despite its rarity, it is of clinical importance, as it may lead to incorrect pre-diagnosis, being confused with other lesions. It is generally seen on the neck and upper extremities. In this case report, pilomatrixoma of the lower extremity has been described, which has been described in the literature extremely rare.

Case Report

A 48-year-old white female patient was admitted to our clinic complaining of right knee pain after a fall, with diffuse tenderness and minimal swelling in the knee joint on physical examination. Range of motion was normal. During the examination, a mobile soft tissue mass of hard consistency, approximately 2x2 cm in the proximal part of the right tibia was determined. There was no discoloration of the skin, or an increase in temperature. The patient reported that the mass was present for three years. The patient had no history of trauma, fever, night sweats, or weight loss. Physical examination revealed no other abnormality. Taking the location of the mass into consideration, lipoma was considered as the initial diagnosis.

X-ray radiographs of the patient were reported as normal. An ultrasound (US) examination revealed a solid mass of 1.5x2 cm, located under the skin. Computerized Tomography (CT) and Magnetic Resonance Imaging (MRI) examinations were not carried out.

In differential diagnoses, epidermoid/dermoid cysts, calcified lymph nodes, calcified hematomas, foreign bodies, sebaceous cyst-like lesions, and chondroma should be considered.

The mass was totally excised together with the surrounding soft tissue, under local anesthesia (figure 1).

Histological examination of the excised mass was reported as pilomatrixoma. No local recurrence was observed in the 1-year follow-up of the patient.

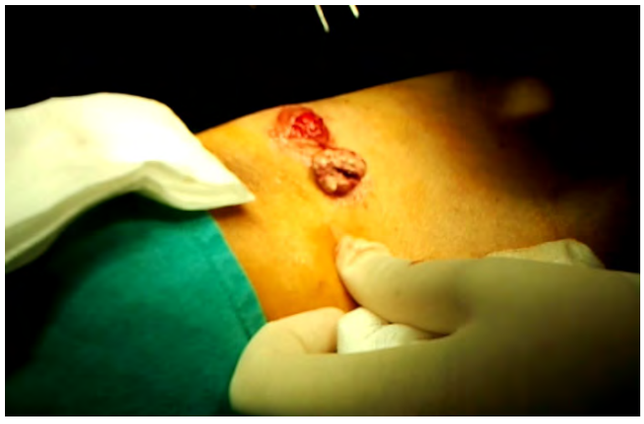

Figure 1. Clinical viewing of excised mass

Discussion

Pilomatrixoma is a benign tumor derived from cells of the hair follicle, which is usually localized in the neck region [1]. The name pilomatrixoma was given by Forbis and Helwig in 1961 [2]. It can be seen at any age and is more common in women [3]. Although its etiology is unknown, it may be associated with trauma, infection, or a pause in the cycle of the hair follicles [4]. Two major parts are characteristic for pilomatricoma: basophilic cells and shadow cells for pathological investigation. Although the genetic mutation has been questioned in some cases, a genetic disorder was not considered in our case.

Pilomatrixomas have been reported often on the trunk, rarely in the upper extremities, and the head and neck region [5]. It has been defined previously extremely rare in the lower extremities. Clinically, the lesion is typically hard as cartilage or bone. The other typical features are slow growth, ease of movement under the skin, and blue-red discoloration of the skin.

In the literature, there are rare malignant cases with distant metastases originating from Pilomatrixoma [6]. Pilomatrix carcinoma is typically painless, often in larger sizes compared to the benign type, and often prone to local recurrence. Metastases are rare. For the diagnosis, examinations such as ultrasound, fine-needle aspiration biopsy, CT, and MRI are used. The rate of correct diagnosis is still not high. Definitive diagnosis requires a histopathological examination. These tests are used for the differential diagnosis of the tumor depending on the localization of the tumor. The differential diagnosis includes epidermoid/dermoid cyst, calcified lymph nodes, calcified hematoma, foreign body, sebaceous cyst-like lesions, and chondroma [7]. In our case, because there were no changes in the size of the lesion for a long time and due to the location of the lesion, no further investigations other than USG were performed. In the intraoperative examination, the mass was approximately 2x1.5 cm in size, hard in consistency, without any discoloration, and mobile in nature. The postoperative histopathological diagnosis was reported as pilomatrixoma (figure 2).

The rate of correct pre-diagnosis of this lesion is very variable. In particular, as the physician is not familiar with such a case, it does not come to mind. Like in pilomatrixoma, in the malignant form, which has a high potential for local recurrence, a resection is required, with removal of 1-2 cm of the surrounding healthy tissue in order to prevent the local relapse [8]. The recurrence rate has been reported as 0-3% in the literature.

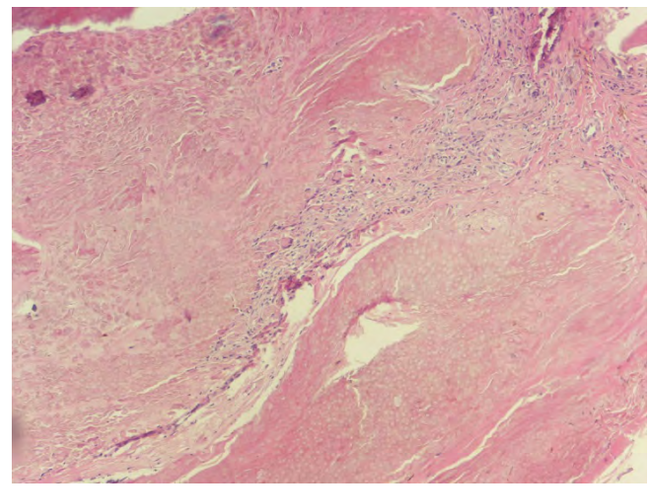

Figure 2. Histopathological image of mass. (HE X 40)

Conclusion

- Pilomatrixoma is a very rare benign soft tissue tumor.
- It can be overlooked, if a careful examination is not carried out.
- Although it is usually seen in neck region, it may also be encountered in other parts of the body.
- It should be totally excised, since there is a slight risk of malignant transformation.
- After the histopathological diagnosis, the patient should be followed-up on for recurrence.

Competing interests

The authors declare that they have no competing interests.

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Case Report
JCAM

Inadvertent Arterial Puncture During Central Venous Catheter Insertion

Santral Ven Kateterizasyonunda Kasıtsız Arter Delinmesi

Arterial Puncture During Central Venous Catheter Insertion

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Özet

Santral ven kateterizasyonun en korkutucu ve tehlikeli komplikasyonlarıdan biri kasıtsız arter delinmesidir. Mevcut çalışmada, bir kasıtsız karotis arter delinme vakiasi santral ven kateterizasyonda bildirilir. Hastamizda hipotansif ve ciddi hipoksemik olmak nedeniyle ve önceki santral ven kateterizasyonunn hasarlı olmak nedeniyle, yeni santral ven kateterizasyonu gerekli oldu. Benzerli durumlarda, santral ven kateterizasyonun doğruluğunu kanatlamk için eşzamanlı oksijen içerik farkını kanüle olan dammar ve arter kinyla ölçmek önerilmektedir.

Anahtar Kelimeler

Santral Ven Kateterizasyon; Hipoksemi; Kasıtsız Arter Delinmesi

Abstract

One of the most important and dangerous complications of central venous catheterization is unintentional arterial puncture. In the current study, were report a case of unintentional arterial catheterization into the carotid artery in a hypotensive patient with severe hypoxemia in which central venous catheterization was required due to loss of intravenous line. In similar situations, it is suggested to measure the difference between O2 content of the cannulated vessel and arterial blood simultaneously for further confirmation of the central venous catheterization.

Keywords

Central Venous Catheter; Hypoxemia; Unintentional Arterial Puncture

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Introduction

Patient safety and acquiring skilled practice has always been of great importance for the physician of all eras [1;2]. Central venous catheters (CVCs) provide us with the evaluations of the hemodynamic parameters which could not be assessed via non-invasive methods. Furthermore, medication administration and nutritional supports which could not be delivered through peripheral lines can be easily given to patients using CVCs [3;4]. Nevertheless, administration of CVCs has been reported to be associated with undesirable complications imposing extra costs to both patients and health systems. More than 15% of the patients with CVCs have suffered numerous complications including mechanical (5-19%), infectious (5-26%) and thrombotic (2-26%) complications [3].

Case Report

Our patient was a 78 year old male who had undergone mechanical ventilation due to acute respiratory distress syndrome (ARDS) and refractory septic shock. His hemodynamic status was monitored continuously through the existing arterial line and hit vital signs were as follows: SBP=55mmhg, DBP=24mmhg, Pao2=45mmhg, PH=6.98, Lactate›15mg/dL, Paco2=12mmhg, HCO3=4meq/lit

Due to loss of the existing right Internal jugular CVC and the prompt need for establishing an appropriate intravenous line, it was decided to establish a CVC through left internal jugular vein. To confirm the correct placement of the catheter, it was attached to a pressure transducer which did not show any pulsatile or forward-coming blood stream or any high pressure flow of blood into the line. Furthermore, taken samples from the CVC were dark similar to venous blood. Considering the critical status of the patient and the existing evidence, to increase the blood pressure of the patient noradrenaline infusion was initiated from the established CVC. After a few minutes, commencement of a pulsatile and forward-coming blood stream with high pressure flow of blood into the line assured us from the incorrect placement of the CVC in the artery.

Discussion

CVCs have been reported to be associated with numerous complications including catheter-related infections, thrombosis, pneumothorax, arterial puncture, incorrect placement, chylothorax, hydrothorax, right atrium puncture and airway compression due to hematoma [3].

Pneumothorax, arterial puncture and hematoma are of the most common mechanical complications of CVCs [3;5]. Pneumothorax and hemothorax are more common in subclavian compared to internal jugular approach whereas arterial puncture is more common in internal jugular approach. Arterial puncture and hematoma are also common in femoral approach; however, subclavian and jugular approaches are mostly preferred to femoral approach due to its more frequent association with mechanical complications [3;4]. Arterial puncture has been reported to occur in 6.3-9.4% of the jugular approach, 3.1-4.9% of the subclavian approach and 9-15% of the femoral approach [3]. Factors contributing to increased complication rates in the patients include inexperienced intervener, increased number of attempts for catheterization, 20>BMI>30, short neck, low blood pressure, coagulopathy, large size catheter (e.g. dialysis catheter), history of radiotherapy on the site, previous catheterization and catheterization in the emergency situations [4;5].

Different methods have been introduced to increase the success rate of establishing CVCs as following:

1. Experienced intervener: similar to most medical procedures, the rate of the complications decreases according to the expertise of interveners[3].
2. Ultrasonography has gained its way through all fields of the medicine [6]. The use of ultrasonography guide throughout the catheterization: this would decrease complication rates. Using ultrasonography guide throughout the internal jugular catheterization leads to decreased in mechanical complications. However, administration of ultrasonography guide throughout the subclavian vein catheterization has been associated with controversial results due to its anatomical difference. The needle of the catheter is echogenic in ultrasonography while vascular structures can be seen as echoless (black) areas. Key characteristics in ultrasonography assist us in differentiating vein from artery as veins could be easily compressed and they have thin walls without arterial pulsation. In the hospital and medical centers equipped with ultrasonography facilities, physicians should gain the required expertise and utilization of ultrasonography should be borne in mind especially in internal jugular vein catheterization [3-7].
3. Anatomical landmarks: using the landmarks in normal anatomical conditions without normal variations could decrease complication rates [4;7].

Approaches used for confirming that correct placement of the catheter include:

1. CXR: Performing CXR following catheterization is essential to rule out probable pneumothorax and haemothorax and to confirm the position of the catheter tip. CXR should be taken in standing or semi-sitting positions for a better evaluation of air or liquid existence. In ill patients, patient rotation or oblique ray emission could mislead the physician regarding the position of the catheter tip [7].
2. ABG and Po2 content of the catheter sample: following catheter establishment, instantly a blood sample should be sent for ABG and Po2 analysis. This would help us in differentiating arterial from venous blood [4].
3. Attachment of the catheter end to the pressure transducer and evaluation of the pressure curves: this method helps to find the correct place of the catheter. Arterial blood pressure and central venous pressure (CVP) waveforms cannot differentiate arterial and venous placement of the catheter whenever the blood pressure or effective circulatory volume is low [6-8].
4. Evaluation of the blood stream regarding its being pulsatile or not and its colour: in patients with normal blood pressure and arterial oxygen pressure, it would be easy to distinguish arterial puncture due to the fact that following arterial puncture, a pulsatile light red stream of blood could be observed. However, in patients with very low blood pressure, low levels of haemoglobin or low arterial oxygen saturation these findings may not be present. In case of any doubt regarding the introducer needle being either inside an artery or a vein, a single-lumen catheter 18 F should be placed inside the vessel through the wire. This catheter could be attached to a pressure transducer to confirm

the vein wave curves [4;6].

In our patient, considering his being ill and conditions such as low blood pressure and hypoxemia due to underlying ARDS, he could not be sure of the correct catheter placement using CXR due to rotation; or ultrasonography due to lack of arterial pulse; or CVP curves due to low blood pressure; or the color or the pulse of the bloodstream due to our patient's being hypoxemic and hypotensive. In similar conditions, considering the fact that our patient already had an arterial line, we could confirm the correct placement of the catheter comparing Po2 levels of the samples taken from the catheter and the arterial line. However, in patients without arterial line, being hypoxemic and or hypotensive would also limit us regarding the samples taken from peripheral vessels. This leads to the question that how the correct catheter placement could be confirmed in cases without arterial line.

Competing interests

The authors declare that they have no competing interests.

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Cutaneous Metastasis of Large Cell Lung Cancer: A Case Report

Büyük Hücreli Akciğer Kanserinin Cilt Metastazları: Olgu Sunumu

Cutaneous Metastasis

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Özet

Akciğer kanseri dünya genelinde yıllık insidansı en fazla olan kanser türüdür. Cilt, akciğer kanserinin metastaz yaptığı nadir organlardan biri olup insidansının %1 ile %12 arasında değişiklik gösterdiği bildirilmiştir. Bu makalede 67 yaşında yaklaşık 2 aydır vücudunun çeşitli bölgelerinde şişlik yakınmalarıyla başvurduğu hekim tarafından çekilen akciğer grafisi sonrası, sağ üst zonda yaklaşık 3 cm çaplı homojen dansite artışı saptanması nedeniyle hastanemizin iç hastalıkları polikliniğine başvuran ve sonrasında tarafımıza konsülte edilen bir erkek olgu sunuldu. Özgeçmişinde 80 paket/yıl sigara içtiği öğrenilen hastanın yapılan fizik muayenesinde vücudunun çeşitli bölgelerinde çok sayıda deri lezyonu saptandı. Sağ 6. interkostal aralık ile orta aksiller hat kesişim yerindeki yaklaşık 1,5x1 cm' lik cilt nodülü gerekli hazırlıkları takiben lokal anestezi altında bütün olarak eksize edildi ve patoloji laboratuvarına gönderildi. Patoloji raporunda "büyük hücreli nöroendokrin karsinom" olarak tanısı konuldu ve başka bir odak saptanmaması sonucu akciğerin büyük hücreli karsinomunun uzak metastazı olarak kabul edildi. İnoperabl olarak değerlendirilen hasta onkoloji polikliniğine yönlendirildi.

Anahtar Kelimeler

Deri Metastazı; Akciğer Kanseri; Büyük Hücreli Kanser

Abstract

Lung cancer has the highest incidence among all cancer types in the world. Skin is an uncommon organ that lung cancers metastasize and the incidence of cutaneous metastasis has been reported between 1-12%. In this report, we would like to present the case of a 67 year old male patient who admitted to our hospital with the complaint of multiple swollen masses on the different parts of his skin and has a homogenous mass with the width of 3 cm on chest x ray. The nodule at the intersection of the right 6th intercostal space and the mid-axillary line and with the dimensions of 1.5x1 cm was excised under local anesthesia and the specimen was sent to the pathology laboratory for histopathological examination. The diagnosis of "large cell neuroendocrine carcinoma" was made histopathologically. The patient was diagnosed as the distant metastasis of the large cell lung cancer, considered inoperable and referred to oncology clinics.

Keywords

Lung Neoplasms; Skin Metastasis; Carcinoma, Large Cell

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Introduction

Lung cancer has the highest incidence among all cancer types in the world [1]. Skin is an uncommon organ that lung cancers metastasize and the incidence of cutaneous metastasis has been reported between 1-12% [2]. These metastasis can present themselves to the physician as painless subcutaneous or intramuscular masses that erode the tissues above and become chronic cutaneous ulcers. In this report, we would like to present the case of patient that was diagnosed as the cutaneous metastasis of the large cell lung cancer without the risks of the open biopsy techniques and emphasize the importance of a thorough physical examination including the cutaneous system.

Case Report

A 67 year old male patient admitted to our hospital after admitting to another physician with the complaint of multiple swollen masses on the different parts of his skin and has a homogenous mass with the width of 3 cm on chest x ray. In the past medical history he had an 80 packs/year of smoking and in the physical examination there were multiple, hard and immobile subcutaneous nodules at suprapubical area, left suprascapular area, the intersection of the right 6th intercostal space with the mid-axillary line, and left upper quadrant of the abdomen with the dimensions of 3x2 cm, 5x4 cm, 1.5x1 cm and 1x1 cm respectively (Figure 1). Respiratory system examination revealed

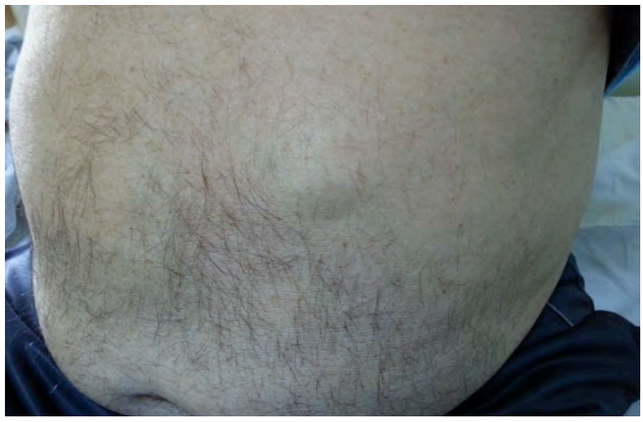

Figure 1. Subcutaneous nodule at the left upper quadrant of the abdomen.

bilateral softened breath sounds. Cervical, supraclavicular and axillary lymph node examinations revealed no lymphadenopathies. Thoracic computerized tomography (CT) was performed and revealed a mass in the posterior segment of the right upper lobe with the dimensions of 4x3 cm and multiple subcutaneous nodules (Figure 2). Patient was hospitalized to the thoracic surgery clinics for taking excisional biopsy from one of his cutaneous lesions. After making the necessary preparations for the surgery, the nodule that was localized at the intersection of the right 6th intercostal space and the mid-axillary line and with the dimensions of 1.5x1 cm was excised under local anesthesia and the specimen was sent to the pathology laboratory for histopathological examination. Immunohistochemical stains of cytokeratin 7 (CK7, Thermo Scientific®), CK20 (Genemed®), epithelial membrane antigen (EMA, Dako), thyroid transcription factor 1 (TTF1, Dako®), synaptophysin (Thermo Scientific®), chromogranin (Genemed®), pankeratin (Thermo Scientific®), high molecular weight cytokeratin (HMWCK, Genemed®) and

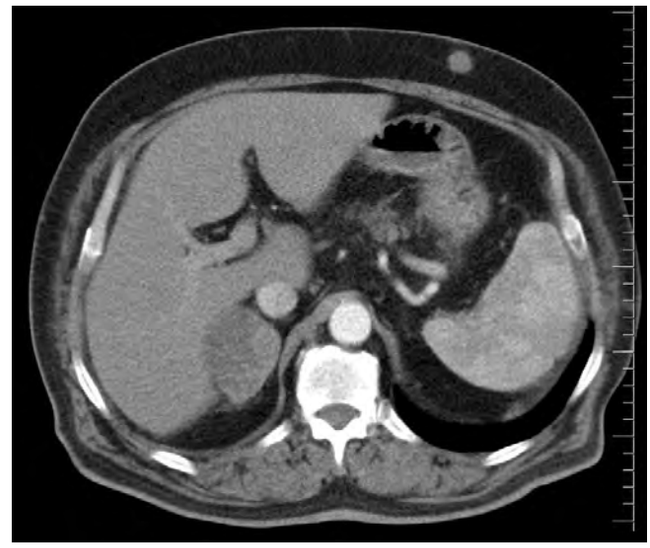

Figure 2. CT section of the subcutaneous nodule at the intersection of the right 6th intercostal space with the mid-axillary line with dimensions of 1.5x1 cm.

tumor protein 63 (p63, Dako®) were tested. Immunohistochemical stains were performed via the automatic immune staining machine (Leica-Bondmax®). The slides were evaluated at a light microscope (Carl Zeiss Axioscope® Photomicroscope) and photographs were taken with an onboard camera system (Carl Zeiss Axiocam ICc3® 3.3 Mp digital camera and Carl Zeiss Axiovision Software®). A tumor that was formed of groups of cells with organoid and trabecular pattern, which includes many atypical mitoses and large necrotic areas, was found in the histopathological examination. Strongly positive immune staining with Pankeratin, EMA, TTF1, CK7 and weakly positive immune staning with synaptophysin and chromogranin. The staining pattern of pankeratin was diffuse not dot-like. There was no immune staining with CK20, p63 and HMWCK (Figure 3). Basaloid type squamous cell carcinoma was eliminated with HMWCK

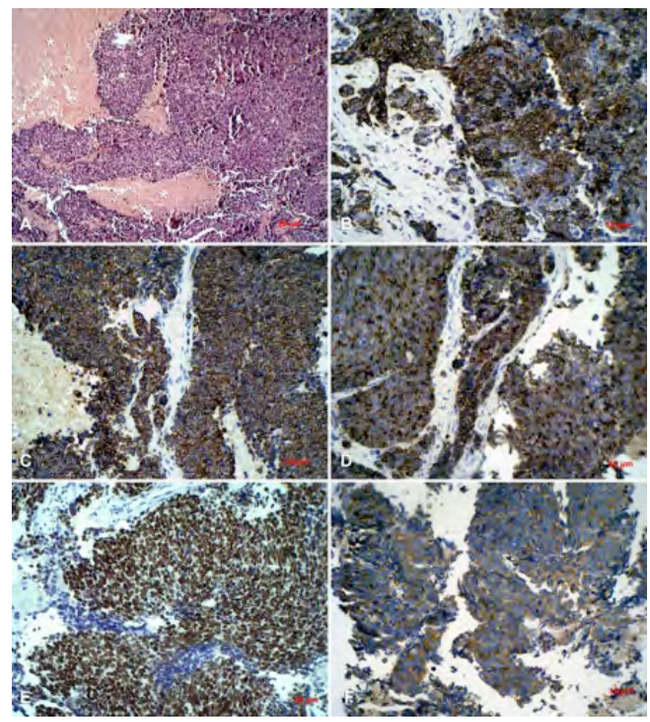

Figure 3. Picture A: Tumor with large necrotic areas that is composed of large cells with hyperchromatic nuclei (HEx100), Immunohistochemical stains: B: CK7 positivity (x200), C: Pankeratin positivity (x200) D:EMA positivity (x200), E:TTF1 positivity (x200), F: sinaptofizin positivity (x200).

and p63 negativity. The diagnosis of "large cell neuroendocrine carcinoma" was reported when histopathological findings were considered along with immunohistochemical findings. TTF1 can also be expressed in extrapulmonary tumors such as thyroid, bladder and prostate along with lungs. The primary pulmonary malignancy was considered in our patient since there was no other focus of malignancy. Patient was discharged on the same day because his postoperative reexamination revealed no abnormalities. The patient was diagnosed as the distant metastasis of the large cell lung cancer. He was considered inoperable and referred to oncology clinics.

Discussion

Cutaneous metastasis of lung cancer was reported more frequently in men than women [2,3,4]. Especially in obese patients subcutaneous nodules are thought of benign origin such as lipomas as in the case of our patient and usually neglected. Cutaneous metastasis, which is rarely seen in lung cancer, can be undiagnosed and thus advanced stage diseases are underrated and unnecessary thoracic surgeries are performed. Several studies indicate that adenocarcinoma of the lung is the most common type of lung cancer that metastasize to skin. But it is also emphasized in other studies that large cell cancer has a higher tendency to metastasize to skin than other types of lung cancer despite the fact that it is a less frequently seen type of lung cancer [2,3]. The cutaneous nodules of our patient that were excised was reported as "metastasis of large cell carcinoma" with histopathological examination. The cutaneous metastasis of the lung cancer was reported to be frequently localized at the head and neck region, thorax and abdomen [4]. Cutaneous nodules were found at the neck, thorax and abdomen of our patient. He also had a nodule at the inguinal region.

The expected mean survival time in a patient with cutaneous metastasis of any cancer type is 6.5 months. But it is reported as low as 2.9 months in cutaneous metastasis of lung cancer [5]. Lung cancer with skin metastasis is considered as inoperable because of these facts. Chemotherapy, radiotherapy or the combination of these two entities can be the treatment of choice in patients with skin metastasis. But despite these treatments achieving complete remission is usually not possible [6]. Biopsy methods can vary according to their level of invasiveness from the trans-thoracic fine needle aspiration biopsy to the biopsies taken via thoracotomy may cause a lot of complications in patients especially with short life expectancies such as lung cancer with cutaneous metastasis. Complications like pneumothorax, hemothorax and hemorrhage will compromise the general condition of the patient and delay the necessary treatments such as chemo-radiotherapy. Physical examination must be thorough and consist of all systems in the body because of these possible complications. Finding a suspicious skin nodule, which can easily be undiagnosed during the physical examination, can prevent unnecessary biopsy operations. But the most important thing in these kinds of situations is performing an unnecessary operation to the patient because of an undiagnosed skin metastasis. Preventing this big mistake can be prevented by a thorough physical examination.

In conclusion, a thorough physical examination should be performed in a patient with an intensive smoking history and cutaneous metastasis of the lung cancer should be suspected if early onset skin nodules are found in the dermatological examination.

Competing interests

The authors declare that they have no competing interests.

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Intrathoracic Schwannoma Presented with Hemothorax

Hemotoraksa Sebep olan İntratorasik Schwannom

Schwannoma and Hemothorax

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Bu olgu sunumu Toraks Derneği 15. Yıllık Kongresinde (Nisan 2012 - Antalya) poster bildirisi olarak sunulmuştur.

Özet

İntratorasik schwannomalar nöral kılıfta bulunan Schwann hücrelerinden köken alan benign ve asemptomatik tümörlerdir. İntratorasik schwannomaların spontan hemoraji sonucu hemotoraksa yol açması nadir görülen bulgudur. Nefes darlığı ve sağ kolda uyuşma şikayetleri bulunan 17 yaşındaki bayan hastaya çekilen ön-arka akciğer grafisinde sağ hemitoraksta masif plevral efüzyon saptanmış olup yapılan torasentezde hemotoraks ile uyumlu sıvı alınması üzerine tüp torakostomi uygulandı. Çekilen kontrol ön-arka akciğer grafisinde sağ hemitoraksta kitle tespit edildi, bilgisayarlı toraks tomografisinde interkostal sinirlerden köken alan 76 x 104 mm boyutlarında kitle tespit edildi. Rezeke edilen kitlenin histopatolojik incelemesinde schwannoma olduğu raporlanan hasta rezeksiyon sonrası on dört aydır hastalıksız hayatına devam etmektedir.

Anahtar Kelimeler

İntratorasik Schwannom; Hemotoraks; Torakotomi; İnterkostal

Abstract

Intrathoracic schwannomas are typically benign and asymptomatic tumors that originate from the Schwann cells of a neural sheath. Hemorrhage from intrathoracic schwannomas is an uncommon finding. We present the case of a 17 year-old girl who had dyspnea and numbness of the right arm. Chest x–ray showed a right-sided massive pleural effusion and exploratory puncture showed hemothorax. After tube thoracostomy and drainage of the bloody effusion, a mass was observed in the right upper hemithorax. Chest tomography revealed a 76x104 mm mass arising from the fourth intercostal nerve. The tumor was successfully resected and, on histopathologic examination it was reported as a schwannoma. After resection, the patient has remained problem-free for fourteen months.

Keywords

Intrathoracic Schwannoma; Hemothorax; Thoracotomy; Intercostal

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Introduction

Neurogenic tumors are commonly found, especially in the posterior mediastinum or in the chest wall, and have a variety of clinical features [1]. Intrathoracic schwannomas are typically benign and asymptomatic tumors that originate from the Schwann cells of a neural sheath [1,2]. Hemorrhage from intrathoracic neurogenic tumors is extremely rare [3]. A review of the English language medical literature found only 7 previous case reports describing hemothorax due to intrathoracic schwannoma.

Case Report

A 17 year-old girl was admitted to our hospital with dyspnea and numbness of the right arm. She had these complaints for a period of fifteen days. On physical examination there was no breathing sounds in the right hemithorax; there was no neurological malfunction except numbness of the right arm. Chest radiography revealed a massive, right-sided pleural effusion and thoracentesis was performed. Exploratory puncture of the right thoracic cavity revealed a bloody pleural effusion and right tube thoracostomy was performed on the patient due to hemothorax. Following the drainage of 750 cc hemothorax, a second chest x-ray was performed, which showed a smooth, rounded mass on the upper-right part of hemithorax (Figure 1a). We planned a thoracic computerized tomography (CT), which revealed a regular, round mass (86x73 mm) arising from the right thoracic wall, which appeared to displace the trachea to the left side of the mediastinum (Figure 1b). Those details of the tumor

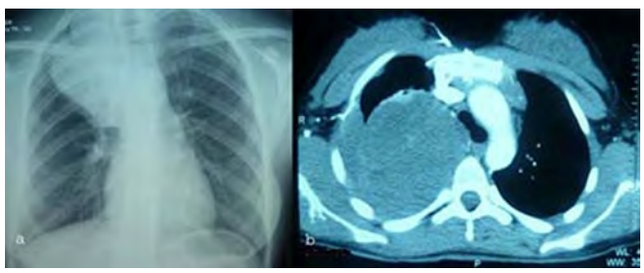

Figure 1. Chest x-ray and thorax computerized tomography revealed smooth, rounded mass on the upper-right part of hemithorax (A, B)

were obscured by the pleural effusion. A decision of transthoracic fine-needle aspiration biopsy was made and pathological examination of specimens indicated a mesenchymal tumor. Hence, we performed chest magnetic resonance imaging (MRI) to clarify the location and characteristics of the tumor. On examination of the chest MRI a huge tumor mass (74x76x104 mm) with high signal intensities was determined on T2-weighted images. On positron emission tomography (PET) examination, the mass had a maximal standard uptake value of 7.26 without lymph node involvement, thus a decision was made to perform thoracotomy for excision procedure.

We performed a posterolateral thoracotomy through the fifth intercostal space and excised the tumor (Figure 2a). In the pathologic examination firm, encapsulated tumor with degenerative changes of both perivascular hyalinization and xanthogranulamatous infiltration was observed (Figure 2b). In light of those findings, a diagnosis of schwannoma was made without difficulty. After the operation she had an uneventful recovery, and has been problem-free without any neurological symptoms for fourteen months.

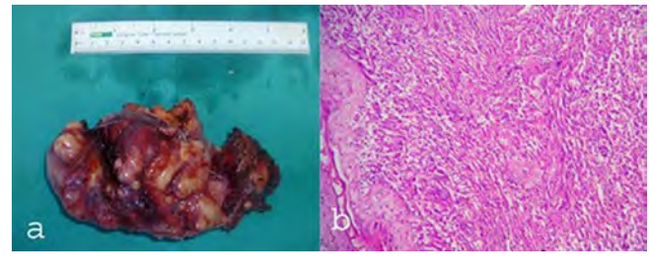

Figure 2. Macroscopic examination of the mass after the resection (A), Microscopic examination of the schwannoma with degenerative changes of both perivascular hyalinization and xanthogranulamatous infiltration (B).

Discussion

Both somatic and autonomic nervous systems are found throughout the thorax and concentrated in the paravertebral sulcus region, known as the posterior mediastinum [1]. Most of the peripheral nerve tumors in the thorax are located in the posterior mediastinum, and 12 % to 21 % of all mediastinal tumors are neurogenic [1]. Intrathoracic schwannomas are uncommon tumors that originate from the Schwann cells of a neural sheath.

In general, schwannomas are slow-growing and asymptomatic tumors except for the compression of neighboring structures [4]. They rarely develop into malignant tumors and the risk of malignancy in a nerve sheath tumor is very small (2% - 5%) [1,2]. On analyzing a series of neurogenic tumors of the chest, Yamaguchi et al. [1] reported that only one (1.7%) of 60 patients had malignant schwannoma. If the patient has a history of radiation exposure or Von Recklinghausen's disease, the malignancy risk increases to 10% - 20% [2]. Malignancy is suggested by features such as mitotic activity, necrosis, nuclear pleomorphism and invasion of surrounding tissues and vascular structures [2]. In our case, there were no malign features and the diagnosis of schwannoma was made without difficulty.

The majority of intrathoracic neurogenic tumors are asymptomatic [1,4]. However, as they become larger in size, these space-occupying lesions can produce local compression symptoms in adjacent structures such as bone erosion and spinal cord involvement [1]. Schwannomas rarely manifest with hemothorax. White et al. [5] reviewed 57 schwannoma cases, of which only 3 were in the chest wall and none presented with hemothorax. In a review the English language medical literature, there were only 7 cases of intrathoracic schwannoma appeared with hemothorax [2,3]. Both Morimoto et al. [2] and Lee et al. [3] described intrathoracic schwannoma cases presented with hemothorax and treated successfully by surgical excision. The causes of bloody pleural effusion were reasoned as bleeding caused by external trauma or a weak locus within the tumor [2]. In our patient, there was no history of trauma; thus, we suspected that some weak locus within the tumor might have been ruptured and caused hemothorax and shortness of breathing.

Typically, schwannomas show equal or lower signal intensities than muscles on T1-weighted MR images. Moreover, they show inhomogeneous high signal intensities on T2-weighted MR images [2]. Morimoto et al. [2] reported a similar case of schwannoma presented with hemothorax and they emphasized the importance of thoracic MRI in preoperative evaluation. Our case also had similar MRI findings to those described in the literature.

Thoracotomy was necessary not only as a diagnostic procedure, but also to ensure completeness of excision and to free a trapped lung [3]. In an analysis of 60 cases, open surgery, including thoracotomy and sternotomy, was performed in 51 patients (85 %), whereas only 9 patients (15 %) were treated using video-assisted thoracic surgery [1]. It was emphasized that best approach would be standard posterior thoracotomy, located one or two intercostal spaces above or below the tumor [1]. In our case, our approach was also standard posterior thoracotomy, one costal space below the mass.

Intraspinal extension of neurogenic tumors requires a combined posterior spinal and thoracic approach for safe resection [6]. A single-stage procedure is recommended because thoracic manipulation of the tumor can produce bleeding within the tumor; hemorrhagic expansion of the tumor within the fixed space of the spinal canal can result in cord compression and paralysis [6]. We performed our case with the help of neurosurgeons although the tumor was not extended to the intraspinal part of the spinal cord. Complete excision of benign schwannomas is considered curative and reported recurrence is uncommon [4].

Conclusion

In conclusion, we report a rare case of intrathoracic schwannoma, originating from intercostal nerve, presenting with hemothorax. Immediate drainage and complete surgical excision of the tumor successfully restored normal pulmonary function. In hemothorax patients a possible underlying neoplasm should always be considered. Consequently, family physicians, neurologists, and thoracic surgeons should be aware that spontaneous massive hemothorax in a patient may result from neurogenic tumors such as schwannoma.

Competing interests

The authors declare that they have no competing interests.

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Retroperitoneal Pararenal Mass; Castleman Disease: A Case Report

Retroperitoneal Pararenal Kitle; Castleman Hastalığı: Olgu Sunumu

Retroperitoneal Castleman Disease

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Özet

Castleman hastalığı lenfadenopati ile seyreden etyolojisi bilinmeyen lenfoproliferatif bozukluktur. Castleman Hastalığı lenfatik zincir boyunca herhangi bir yerde ortaya çıkabilmesine rağmen, mediasten (% 70) en sık bulunduğu lokalizasyondur. Biz abdominal tomografide sağ böbrek renal hilus ile ilişkili homojen retroperitoneal kitlesi olan 36 yaşındaki erkek hastanın olgu sunumunu yaptık. Bu kitleye cerrahi rezeksiyon uygulandı ve rezeke edilen dokunun histopatolojik tanısı Castleman hastalığının hiyalin-vasküler tipi idi. Bu hastalık histolojik ve prognostik olarak malign lenf nodu hiperplazisinden farklıdır. Castleman hastalığı nadir görülen bir durum olmasına karşın, her zaman retroperitoneal tümörlerin ayırıcı tanısında akılda tutulması gereken bir durumdur.

Anahtar Kelimeler

Lenf Nodu; Retroperiton; Bening Tümör

Abstract

Castleman's disease is a heterogeneous group of lymphoproliferative disorders with unknown etiology presenting with lymphadenopathy. Although Castleman's Disease may occur anywhere along the lymphatic chain, the mediastinum is the most common location (70%). We represent 36-year-old male patient with homogeneous retroperitoneal mass that interrelated with renal hilum of the right kidney in abdominal tomography. Surgical complete resection performed and histopathological diagnosis of the resected tissue was hyaline-vascular type of Castleman's disease It is histologically and prognostically distinct from malignant lymph-node hyperplasia. Although Castleman's disease is rare condition, it should always be kept in mind in the differential diagnosis of retroperitoneal tumors.

Keywords

Lymph Node; Retroperitoneum; Benign Tumor

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Introduction

Benign retroperitoneal tumors are rare conditions, comprising only about 20% of all primary retroperitoneal neoplasm's [1]. Castleman's disease is a heterogeneous group of lymphoproliferative disorders with unknown etiology presenting with lymphadenopathy. Although Castleman's disease may occur anywhere along the lymphatic chain, the mediastinum is the most common location (70%). Extrathoracic sites such as the neck, axilla, pelvis, and retroperitoneum have been reported less frequently [2]. It is histologically and prognostically distinct from malignant lymph-node hyperplasia. We present a rare case of unicentric Castleman's disease of the hyaline-vascular type located at right retroperitoneal area contract with kidney vessels.

Case Report

A 36-year-old male patient with one-month history of right flank pain and dyspepsia admitted our clinic. Physical examination, routine hematologic, blood biochemistry and, urine analysis were normal. Abdominal x-ray was considered normal but abdominal ultrasonography demonstrated retroperitoneal mass, with 60 mm in size and with regular contour in the right anterior pararenal space. Then abdominal computed tomography performed and tomography demonstrated 62x60 mm sized homogeneous retroperitoneal mass that interrelated with renal hilum of the right kidney (Figure 1). Testicular examination and testicular ultrasonography were normal. Through a midline abdominal incision, the mass was found near the hilum of the right kidney and it was in contract with the kidney vessels. We performed complete resection of the mass. Patient had no complication in the postoperative period. Histopathological diagnosis of the resected tissue was hyaline-vascular type of Castleman's disease (Figure 2). Postoperative 1 year abdominal tomography was normal.

Discussion

Castleman's disease was first reported in 1956 by Castleman et al. as a different mediastinal mass, easily confused with the thymoma [3]. Castleman's disease is relatively rare and poorly understood lymphoproliferative disorder. Hypotheses for the pathogenesis of Castleman's disease include infection, autoimmunity and dysregulated cytokine expression causing lymphoid proliferation [4]. Mediastinum (70%) is the main location of the disease however retroperitoneal location has been reported in 7%, with only 2% involving the pararenal region [2]. Castleman's disease has no predilection for either sex and affects varying ages of patients but most patients are in the second and third decades of life [5]. Three basic histopathologic subtypes have been described: hyaline-vascular, plasma cell, and mixed variant [6]. Two clinical entities have also been described: a unicentric presentation with disease confined to a single anatomic lymph node, and a multicentric presentation characterized by generalized lymphadenopathy and a more aggressive clinical course [6]. Patients with localized hyalinevascular type are usually asymptomatic, as in our case. Although CT is helpful for the diagnosis of Castleman's Disease, the final diagnosis depends on pathologic examination. The standard therapy for localized, hyaline-vascular form of Castleman's disease is surgical excision, which is curative when resection is complete and en-block; the 5 years of survival is nearly 100%, and no recurrences have been reported [7].

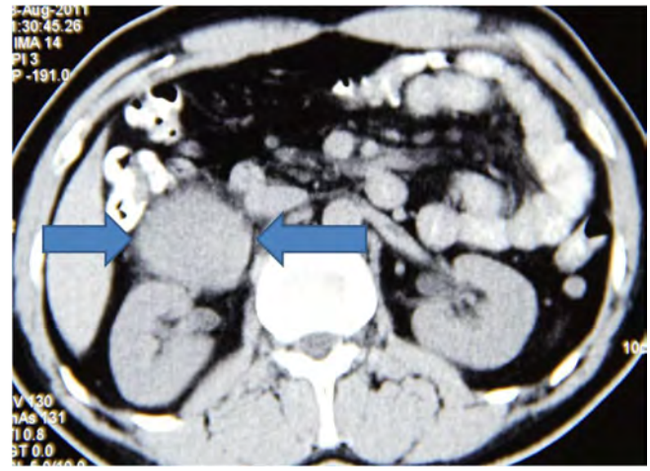

Figure 1. Abdominal tomography demonstrated 62x60 mm sized homogeneous retroperitoneal mass that interrelated with renal hilum of the right kidney

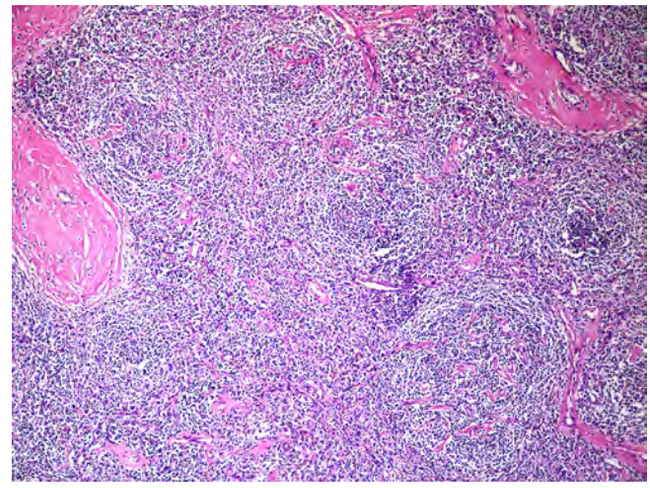

Figure 2. Regressed follicles consist of small germinal centers with marked vascular proliferation and hyaline material. The interfollicular stroma shows hyperplastic vessels and mixed infiltrate of plasma cells, lymphocytes, eosinophils, immunoblasts, and plasmacytoid monocytes (HEX100).

In conclusion, an asymptomatic retroperitoneal mass in a young adult always raises the suspicion of a malignant tumor, but it is necessary to consider hyaline-vascular type Castleman's Disease prominently in the differential diagnosis.

Competing interests

The authors declare that they have no competing interests.

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Case Report
JCAM

Cutaneous Anthrax: Evaluation of Five Family Members

Deri Şarbonu: 5 Aile Bireyinin Değerlendirilmesi

Antrax

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Özet

Şarbon esas olarak sığır, koyun, keçi gibi ot yiyen hayvanların hastalığı olup, insanlara enfekte hayvanlardan bulaşan bir zoonozdur. Her geçen gün azalmakla birlikte ülkemiz için halen endemik bir hastalıktır. Olguların %95'ini deri şarbonu oluşturmaktadır. Bu çalışmada; acil servise el üstünde ağrısız ve siyah renkli yara şikâyeti ile başvurup deri şarbonu tanısı konulan aynı aileden 5 olgu tartışılmıştır. Bir hafta önce büyük baş bir hayvanı kesen 4'ü erkek 1'i kadın aile bireyleri, el sırtında yara ile acil serviste değerlendirildi. Şarbon tanısı konulan hastalardan 1'i enfeksiyon hastalıklarına yatırılarak, 4'ü ayaktan tedavileri düzenlenerek taburcu edildi. Hastaların iki hafta sonraki kontrollerinde her hangi bir komplikasyon gelişmeden tamamen iyileştikleri gözlendi. Erken tanı ve tedavi ile %100 iyileşme gösterebilen deri şarbonu, şüpheli cilt lezyonu ve yakın zamanda hayvan teması olan hastalarda mutlaka düşünülmelidir.

Anahtar Kelimeler

Deri Şarbonu; Cilt Bulgusu; Acil Servis

Abstract

Anthrax is a zoonotic disease of grass-eating animals such as cattle, sheep, goats, and transmitted to humans by infected animals. Declining with each passing day in our country is still an endemic disease. Cutaneous anthrax is 95% of the cases. In this study, 5 patients from the same family who admitted to the emergency department with a complaint of a painless and black colored wound on their hands are discussed and skin anthrax diagnosed. Four male, 1 female family members who cut a cattle a week before were evaluated in the emergency department with a wound on their hands' backside. One of the patients hospitalized with the diagnosis of anthrax, and accepted to the infectious diseases clinic, and 4 were discharged by outpatient treatment arranged. In controls after two weeks, the patients were fully recovered without complications. Early diagnosis and treatment can show 100% improvement in skin anthrax, suspicious skin lesion and recently animal contact history should be considered in patients.

Keywords

Cutaneous Anthrax; Cutaneous Finding; Emergency Service

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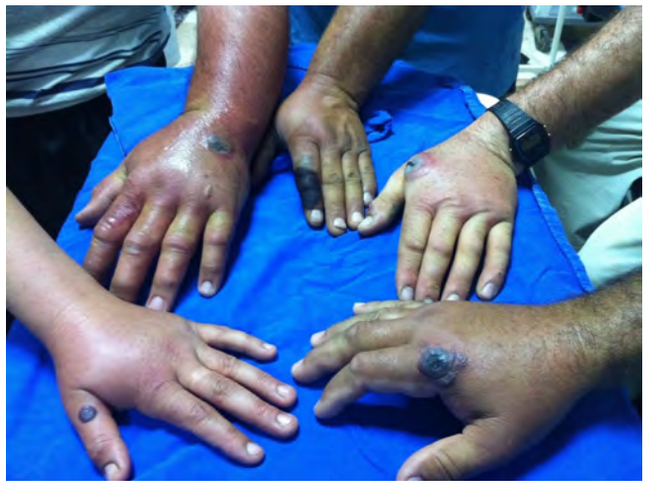
Giriş

Şarbon veya Antraks; Bacillus antracis adlı bakteri tarafından meydana getirilen bulaşıcı bir hastalıktır. Hastalığın bulaşmasında enfekte hayvanın derisi, eti ve yünü ile doğrudan temas önemlidir. İnsana hasta hayvanların kesilmesi ve derisinin yüzülmesi sonucu direkt temasla veya enfekte etlerin yenmesi ya da sporlarının inhale edilmesiyle bulaşmaktadır. Etkenin vücuda giriş yoluna göre deri, barsak ve solu¬num sistemi şarbonu oluşur. Olguların % 95'i deri şarbonu şeklindedir [1]. Deri şarbonu ülkemizde bazı bölgelerde halen endemik olarak görülmesine rağmen son yıllarda sıklığı giderek azalmaktadır.
Deri şarbonu tedavi edilmediğinde yaklaşık %20 oranında mortalite riskine sahiptir [2]. Erken tanı ve tedavi, komplikasyon oluşumunu anlamlı derecede azaltabilir. Bu çalışmada konuya dikkat çekilmesi amacıyla servisimizde tanı konan aynı aileden 5 şarbon olgusu sunulmuştur.

Olgu Sunumu

El üstünde yara şikayeti ile aynı anda başvuran ve akraba olan 5 hastanın yapılan muayenelerinde her bireyin elinin farklı yerinde etrafı ödemli, ağrısız, ülsere siyah lezyon tespit edildi (Resim 1). Hastaların 4'ü erkek 1'i kadındı. Hastalardan alınan hikayede bir hafta önce büyük baş bir hayvanı kesip derisini yüzdükleri ve etlerini parçaladıkları öğrenildi. Hastaların tümünde lezyonların tarifi aynıydı. Acil servise başvurudan 4-5 gün önce lezyonların küçük bir sivilce şeklinde başladığı giderek koyulaştığı ve büyüdüğü ve bu arada ellerinde şişlik oluştuğu öğrenildi. Hastaların özgeçmiş ve soy geçmişlerinde herhangi bir özellik yoktu. Sistemik fizik muayenelerinde patolojik bir bulguya rastlanmadı. Hastalara ait demografik veriler, vital bulgular ve laboratuar sonuçları tablo 1'de özetlenmiştir. İki numaralı hasta kliniğinin ve laboratuar sonuçlarının daha ağır olması sebebiyle enfeksiyon hastalıklarına konsülte edilerek bölüme yatırıldı. Bölümde 2,4 Milyon Ü/gün İV Penisilin G tedavisi alan hasta üç gün sonra enfeksiyon hastalıklarından 800000 Ü 2x1 İM (10 gün) reçete edilerek taburcu edildi. Diğer 4 hasta ise penisilin G 800000 Ü 2x1 İM başlanarak acil servisten taburcu edildi. Hastaların iki hafta sonraki kontrollerinde her hangi bir komplikasyon gelişmeden cilt lezyonlarının tamamen iyileştiği gözlendi.



Resim 1. Beş olgunun başvuru anındaki lezyon görünümleri

Tablo 1. Hastaların demografik verileri, vital bulguları ve laboratuar sonuçları

Hasta No Parametreler	1	2	3	4	5
Yaş (Yıl)	44	49	45	36	33
Cinsiyet	E	E	E	K	E
Solunum/dak.	13	15	13	14	14
Nabız/dak.	91	97	90	88	85
TA (mmHg)	125/60	100/60	150/90	125/68	125/80
Ateş (oC)	38	39	36,5	37,8	36,5
Beyaz Küre (WBC) (/µL)	11400	18600	10000	11500	10600
Nötrofil (%)	67	83,1	67,2	73,7	74,5
Sedimantasyon (mm/s)	25	30	28	28	26
CRP (mg/L)	50	200	80	30	45
Yatış süresi	0	3	0	0	0

Tartışma

Hayvancılığın ve hayvan kesiminin modern yöntemlerle yapılmadığı ülkemizin bazı bölgelerinde bir zoonoz olan şarbon endemik olarak görülmeye devam etmektedir [1]. Ana etken olan Bacillus anthracis gram-pozitif, kapsüllü, sporları toprakta uzun yıllar canlı kalabilen bir bakteridir. Ana bulaş yolu sporlarla direkt temas veya inhalasyon yolu iledir [2]. Ülkemiz için yapılan bir çalışmada olguların % 92'sinin şarbonlu hayvanın kesilmesi, yüzülmesi gibi işlemler sırasında direkt temasla, % 1.5'inin enfekte et yemekle bulaştığı ve % 6 olguda bulaş şeklinin saptanamadığı bildirilmektedir [3]. Bizim çalışmamızdaki 5 vakanın hepsi aynı hayvanın kesimi ve derisinin yüzülmesi işlemine iştirak etmiştir. Bu sebeple bulaşın bu hayvan aracılığı ile olduğunu düşünmekteyiz.
Şarbon genel olarak 3 temel formda karşımıza çıkar. Bunlar deri, solunum yolu ve gastrointestinal şarbondur. Deri şarbonu tüm vakaların %95 ini oluşturur. Bulaşmayı bütünlüğü bozulmuş deri varlığı kolaylaştırır. Deri lezyonları sporların deriye girişini takip eden 2-7 gün içinde kendini gösterir [4]. Sporlar deriden girişi takiben vejatatif şekle geçer ve giriş yerinde hafif yanma ve kaşıntı olur. İki üç gün sonra iltihabi bir papül meydana gelir. Bu papül kısa bir zamanda etrafı eritem ve ödemli, içi serohemorajik sıvı ile dolu bir büle dönüşür. Büller zamanla patlar veya üzerinde siyah renkli krut meydana gelir. Tüm basamaklarda lezyon ağrısızdır [4]. Bizim vakalarımızın hiç birinin ellerinde bulaş öncesinde bir yaralanma yoktu. Vakaların tümünde, lezyonlar hayvan kesiminden 2-3 gün sonra el üstlerinde küçük bir sivilce şeklinde başlamış giderek koyulaşmış, büyümüş ve bu arada ellerinde şişlik oluşmuştu. Bize başvurulduğunda her bireyin elinin farklı yerinde etrafı ödemli, ağrısız, ülsere siyah lezyon tespit edildi.
Şarbonun her üç klinik formu da tedavi edilmediği takdirde öldürücü olabilir. Uygun tedavi edilmeyen vakaların %10-40'ında ölüm gözlenirken, tedavi edilen vakalarda bu oran % 1'e gerilemektedir [4]. Hastalığın tedavisinde penisilin G halen ilk tercih edilen antibiyotiktir. Tetrasiklin, eritromisin ve siprofloksasin özellikle penisilin alerjisi olan vakalarda diğer alternatif tedavi seçenekleridir. Komplike olmamış orta şiddetli şarbonda intramüsküler penisilin önerilirken, artmış ödemin bulunduğu,

sistemik belirtilerin şiddetli olduğu vakalarda intravenöz penisilin tedavisi önerilmektedir [5,6]. Çalışmamızda 2 numaralı vaka enfeksiyon hastalıklarına yatırılarak 3 gün 2,4 Milyon Ü/gün İV Penisilin G tedavisi alıp penisilin G 800000 Ü 2x1 İM tedavisi ile bölümden, diğer 4 hasta ise penisilin G 800000 Ü 2x1 İM tedavisi ile acilden taburcu edildi.

Şarbonda penisilin tedavisinin süresi netlik kazanmamıştır. Genel olarak tedavinin başlamasından 7-10 gün sonra lezyonlar geriler [4,5]. Biz hastalarımızı 10 günlük penisilin tedavisi başlayarak taburcu ettik. İki hafta sonra yapılan kontrolde tüm vakaların komplikasyonsuz iyileşmiş olduğunu gördük.

Sonuç olarak, şarbonun endemik olarak görüldüğü bölgelerde hastalığa bağlı morbidite ve mortalitenin azaltılması için özellikle bu bölgelerde çalışan hekimlerin, şarbonun bulgu ve semptomlarını kolayca tanımaları, erken ve etkili bir tedaviye başlanabilmesi açısından önemlidir. Özellikle hayvanlar ile yakın teması olan ve ağrısız siyah kurutlu lezyon görülen vakalarda deri şarbonu akılda bulundurulmalıdır.

Competing interests

The authors declare that they have no competing interests.

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Heparin-Induced Thrombocytopenia Association with Impaired Liver Function Tests

Bozulmuş Karaciğer Fonksiyon Testleri ile Heparine Bağlı Trombositopeni Birlikteliği

Heparin-Induced Thrombocytopenia

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Özet

Heparine bağlı trombositopeni son derece prokoagülan bir hastalıktır ve önemli morbidite ve mortalite taşımaktadır. Bununla birlikte, serum transaminaz yüksekliği karakteristik olarak asemptomatiktir ve tedavinin sonlandırılması ile tersine çevrilebilir. Heparine bağlı ciddi karaciğer zararı rapor edilmemiştir fakat hepatosit nekrozu ile beraber ciddi karaciğer hasarı potansiyeli olduğu için karaciğer fonksiyonları yakından izlenmelidir. Bu vaka takdiminde, pulmoner arter bantlama prosedürü sonrasında sol alt ekstremitede arteriyel tromboz gelişen, bir yaşında bir erkek hasta sunmaktayız. Olgumuz antikoagülasyon için fraksiyone olmayan heparin almaktaydı. Heparin tedavisinin üçüncü gününde, rutin kan tahlillerinde hepatik fonksiyon testlerinde bozulma ve trombositopeni saptandı. Standart heparin tedavisi kesildi ve arteriyel tromboz terapötik dozda düşük molekül ağırlıklı heparin ile tedavi edildi. Trombosit sayımı ve karaciğer fonksiyon testleri 2 hafta sonra normale geldi.

Anahtar Kelimeler

Heparine Bağlı Trombositopeni; Karaciğer Hasarı; Arteriyel Tromboz; Çocuk

Abstract

Heparin-induced thrombocytopenia is an intensely procoagulant disorder and carries significant morbidity and mortality. However, the elevations in serum aminotransferases are characteristically asymptomatic and reversible with treatment termination. The serious liver injury due to heparins has not been reported but liver function should be monitored closely since potential severe liver damage with hepatocyte necrosis can occur. In this case report, we present a one-year-old male patient who developed a left lower extremity arterial thrombosis following an pulmonary artery banding procedure. Our case was taking unfractionated heparin for anticoagulation. On the third day of heparin therapy, routine blood analysis revealed deranged hepatic function tests and thrombocytopenia. Unfractionated heparin treatment was discontinued and he was treated with therapeutic doses of low-molecular-weight heparin for arterial thrombosis. His platelet count and liver function tests had normalised 2 weeks later.

Keywords

Heparin-Induced Thrombocytopenia; Liver Damage; Arterial Thrombosis; Children

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Introduction

Unfractionated heparin (UFH) is the traditional anticoagulants used for the prevention and treatment of thromboembolic disease. It is an indirect thrombin inhibitor [1]. Heparin-induced elevations in serum aminotransferases is characteristically asymptomatic and reverse with continued treatment. However, heparin-induced thrombocytopenia is a potentially devastating complication of heparin therapy [2,3]. The best of our knowledge, there are no reported articles on cases of concurrent elevations in serum aminotransferases and thrombocytopenia associated with UFH in children and adolescents. In this report, a one-year-old boy presented with elevations in serum aminotransferases and thrombocytopenia which had been reported to be very rarely seen.

Case Report

A one-year-old, 7-kg boy with secundum atrial septal defect, perimembranous ventricular septal defect and severe pulmonary hypertension underwent pulmonary artery banding procedure. He had left femoral arterial and venous catheters. On the third day of surgery, the developed pallor and loss of pulse his left leg and foot.

He was alert, blood pressure was 90/55 mmHg, body temperature was 36,7°C, heart rate was 118 beats/min , respiratory rate was 42 breaths/min and oxygen saturation was 94%. His breath sounds were normal. Thrombus formation was noted observed in the evaluation of echocardiography. Chest radiographs showed mild cardiomegaly without congestion. Color Doppler ultrasonography (CDU) of the symptomatic leg (left leg) demonstrated no significant colour flow within the common femoral artery, popliteal artery, posterior tibial artery, dorsalis pedis artery, whereas spectral Doppler ultrasonography showed minimal flow within these arteries. Left lower extremity venous CDU were normal. Doppler ultrasonography confirmed a left lower extremity arterial thrombosis. Initial laboratory tests revealed a white blood cell count of 7.680 cells/mL, hemoglobin 12.7 g/dl, platelet count of 240.000 cells/mL, International Normalized Ration (INR) value 1.2 (normal range:1–1.5), and an activated partial thromboplastin time (aPTT) value of 35 s (normal range: 20–40 s). Routine blood biochemistry tests were normal. The thrombus was probably associated with arterial catheter. Treatment with UFH was initiated (70 units/kg intravenous bolus, 20 units/kg per hour continuous infusion, goal aPTT is 60–80 s). Three days after the patient's initial admission and first exposure to heparin, his platelet count had dropped from a baseline of 240.000 to 30.000 cells/mL. Platelet counts for the patient are illustrated in Table 1. There were no other possible causes of the thrombocytopenia. The same time, he was detected to have derangement in liver function tests (LFT). Liver function tests are listed in Table 2. No another drugs known to elevate LFT were given before and after cardiac surgery and all other diagnostic evaluations for liver damage, including viral hepatitis tests and were negative. Ultrasound of the liver was normal. Also, the previous liver function tests from treatment of UFH were normal. Unfractionated heparin was then suspected to be the cause of the derangement in LFT and thrombocytopenia. Therefore, it was stopped. Color Doppler ultrasonography of the left lower extremity show that the disorder was continued. Subcutaneous enoxaparin 0.5 mg/kg every 12 hours was commenced. His LFT and thrombocytopenia started to improve five days after UFH was stopped. The patient was treated with subcutaneous enoxaparin for two weeks. Color Doppler ultrasonography of the left lower extremity was normal on the twelve day of therapy enoksiparin. The LFT and platelet count slowly improved and returned to normal over a period of two weeks.

Table 1. Platelet Counts of the Patient

	PLT (cells/mL)
Baseline PLT (cells/mL)	240.000
PLT 3 Days after UFH Treatment	30.000
PLT 24 h after Stopping UFH	42.000
PLT 1 Week after Stopping UFH	97.000
PLT 2 Week after Stopping UFH	281.000

PLT: Platelet counts

Discussion

Central venous and arterial catheters (CVAC) are used in critically ill children and in children with chronic diseases for the administration of fluids, medications, total parenteral nutrition or blood products. But, despite their many benefits, CVACs are not innocuous and are associated with important complications [4]. Arterial and venous thromboembolism are significant because they are difficult to detect, increase the cost of care, and are potentially life-threatening adverse events [5]. The main stays of anticoagulant therapy for children with thrombus are UFH, low molecular weight heparin, and warfarin. Unfractionated heparin is often the anticoagulant of choice in children because of its efficacy, reversibility, ease of monitoring and clinical experience is extensive [6]. Also, we started initially UFH to our patient. The UFH doses are age dependent,with infants (up to 2 months corrected for gestational age) having the highest requirements (average 28 U/kg/h) and children over 1 year of age having lower requirements (average 20U/kg/h) [3,6].

The major complications of UFH therapy are thrombocytopenia,

Table 2. Serial Liver Function of the Patient

LFT	Baseline LFT	LFT 3 Days after UFH Treatment	LFT 24 h after Stopping UFH	LFT 1 Week after Stopping UFH	LFT 2 Week after Stopping UFH
AST (U/L)	32	10600	2466	197	39
ALT (U/L)	46	7580	5140	1446	39
ALP (U/L	101	112			107
GGT (U/L)	61	121	83		64
LDH (U/L)	214	497	304		221
Total bilirubin (mg/dl)	1.2	1.3			1.2

LFT, liver function tests; AST, aspartate transferase; ALT, alanine transferase; ALP, alkaline phosphatase; GGT, glutamyl transpeptidase; LDH, lactate dehydrogenase; UFH, unfractionated heparin.

bleeding, osteoporosis, elevations in serum aminotransferases, skin necrosis and skin lesions [1,7]. Heparin-induced thrombocytopenia (HIT) is a life-threatening, severe, immunological drug reaction that carries a high risk of morbidity and mortality. The frequency is reported to be 2.3% to 3.7% with a 1% to 3% prevalence in children undergoing cardiac surgery with the use of UFH [3]. It is generally suspected in cases of unexplained acute thrombocytopenia; generally more than 50% fall in absolute platelet count from the baseline, in a patient that has been on heparin for 4–14 days. The diagnosis of HIT initially is clinical. The awareness of the syndrome is necessary to suggest HIT in cases of unexplained thrombocytopenia during heparin exposure [3]. We have excluded other causes of thrombocytopenia and the patients was defined as HIT. Drug-induced liver injury is a major concern for the physicians. Almost one thousand medicines used in clinical practice have been shown to induce hepatotoxicity. in medical literature. Several medication are lipophilic substances and their transformation into hydrophilic compounds by the cytochrome P-450 system results in production of toxic metabolites. The necrotic death follows antioxidant consumption and oxidation of intracellular proteins, which determine increased permeability of mitochondrial membranes, loss of potential, decreased ATP synthesis, inhibition of calcium-dependent ATPase, reduced capability to sequester calcium within mitochondria, and membrane bleb formation. The activation of nucleases and energetic participation of mitochondria are the main intracellular mechanisms that lead to apoptosis. Non-parenchymal hepatic cells are inducers of hepatocellular injury and targets for damage [8]. The exact pathogenesis of distorted liver function tests induced by heparin is so far unknown. The serious liver injury due to heparins has not been reported. It is characteristically asymptomatic and reverse with continued treatment [2,9]. Unfractionated heparin-associated hepatotoxicity is generally detected within the first week of therapy and resolves upon discontinuation of the offending agent. In our patient, the aminotransferase elevation was beginning at 3 days after the initiation of dosing UFH treatment and ıt was returning to normal values within 2 week of discontinuing treatment of UFH. But, some studies showed drug-induced toxic centrilobular hepatocellular balloon degeneration in a patient with high liver enzymes (>3000 U/L) [10,11]. Therefore, should be noted that develop toxic liver necrosis in patients with liver enzymes especially more than 3000 U/L.

In conclusion, anticoagulation therapy in the hospital is widespread and many patients will be exposed to heparin at some time during their hospitalization. Unfractionated heparin is used ubiquitously for the treatment and prevention of thrombosis. However, proper medication use requires an understanding of the medication's indications and side effects.

Competing interests

The authors declare that they have no competing interests.

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Total Pancreatic Fracture Due to Blunt Trauma: Report of a Rare Case

Künt Karın Travmasına Bağlı Total Pancreas Kırılması: Nadir Bir Vaka Sunumu

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Özet

Künt karın travmasına bağlı olarak gelişen nadir bir pankreas fraktürü vakasını sunmaktayız. 70 yaşında erkek hasta araç içi trafik kazası geçirmiş, fizik muayene, ultrasonografi ve bilgisayarlı tomografi incelemeleri sonucunda pankres travmasından şüphelenilmiştir. Dalak yaralanması ve portal venin pankreasın hemen altından geçtiği bölgede total pankreatik fraktür tanısı magnetik rezonans kolanjiyopankreatografi ile konuldu. Hasta acilen operasyona alınarak splenektomi ve distal pankreatektomi uygulandı. Sunduğumuz bu vakada çok sık olarak karşımıza çıkmayan künt travmaya bağlı pankreatik fraktürü, komplikasyonlarını ve tanısal ve tedavisel zorlukları tartıştık.

Anahtar Kelimeler

Pankreas; Abdominal Yaralanmalar; Cerrahi Prosedürler

Abstract

A rare case of pancreatic fracture due to blunt trauma was presented. The patient was 70 year old male who had a motor vehicle collision and was suspected a pancreatic trauma due his examinations with ultrasound and computerized tomography. The diagnosis of splenic injury and pancreas body total fracture in the point where the portal vein crosses the pancreatic body was made with the help of magnetic resonance cholangiopancreatography. He was taken to emergency surgery where a splenectomy and a distal pancreatectomy were performed. We represented this infrequent case of pancreatic fracture and its complications after blunt abdominal trauma and discuss the diagnostic and management practices.

Keywords

Pancreas; Abdominal Injuries; Surgical Procedures

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Introduction

Blunt abdominal trauma can cause severe damages in all internal organs. Trauma to the pancreas especially isolated pancreatic trauma is a very uncommon incident in these injuries[1]. Pancreatic fracture due to abdominal blunt trauma reported to be 0.4 per 100 000 population [2] or 1 per 250 000 hospital admissions[3]. The diagnosis of a pancreatic fracture is challenging as the physical signs are often absent and laboratory parameters such as serum amylase are inaccurate for diagnosis[4]. As the diagnosis and the treatment are difficult, a combined morbidity and mortality rate of 50% has been reported[1]. It is critically important to provide early treatment in these patients, therefore; infrequent pancreatic injuries should certainly be considered in the differential diagnosis in blunt trauma cases.

Herein we represented a rare case of pancreatic fracture after blunt abdominal trauma and discuss the diagnostic and management practices.

Case Report

A 70 year old male admitted to the emergency service with severe abdominal pain after having a motor vehicle collision. He stated that he hit his chest and upper abdominal area through the steering wheel hardly during the accident. In his initial physical assessment; he was stable with a blood pressure of 160/80mmHg, heart rate of 96 /min and fever of 36.4 C. He had an abdominal tenderness especially in upper quadrants. A total blood count revealed a white blood cell count of 15000/mm3, hemoglobin of 12.6 g/dl and serum amylase of 800 iu /i. An ultrasound examination in the emergency service was initiated. A large hypo-echogenic area with dimensions of 10x8 cm was observed around the liver and spleen and interpreted as hematoma secondary to solid organ trauma. For further investigation a computed tomography (CT) scan was performed. Axial contrast-enhanced CT scans showed extensive spleen lacerations and a fracture line at the pancreatic body surrounded with fluid and hemorrhage between the pancreatic fragments. Depending on the CT findings magnetic resonance cholangiopancreatography (MRCP) was initiated and axial T2 weighted fat suppressed magnetic resonance (MR) image showed a fracture of the pancreatic body with associated fluid collections. Preoperative diagnosis of an acute abdomen due pancreatic fracture was decided and the patient underwent emergency surgery. In the exploration of the abdomen; a spleen rupture was seen with an intra-abdominal bleeding of approximately 2 liters of blood was suctioned. In addition, the mesentery of small and large intestine was found to be filled with hematoma due pancreatic fracture. A retroperitoneal dissection were performed and found that the pancreas body was fractured totally in the point where the portal vein crosses the pancreatic body. Pancreatic fluid leakage was also inspected. After the exploration; a splenectomy and a distal pancreatectomy were performed. A silicon drainage tube was placed in the resection area of the pancreas before the anatomical closure of the patient.

The patient follow-up postoperatively was uneventful and he was discharged from the hospital on day 4.

After one week of the discharge, he re-admitted to our clinic with severe pain of the abdomen and high fever of 39 C. He had a white blood cell count of 20000/mm3. CT scan examination showed multi centered fluid and abscess like fluid, and therefore the patient underwent relaparotomy. In the intraabdominal exploration; a close small intestine perforation adjacent to the resection area (due pancreatic fluid digestion) were found with intraabdominal multiple abscess foci. The perforated intestinal section was resected, the abscesses were drained as much as possible and the operation was ended with a replacement of silicon drainage. In the first postoperative day, the drainage fluid laboratory assessment showed very high amylase levels and an MRCP was planned. MRCP showed pancreatic fluid leakage from the resection area. An ERCP-guided stent placement is considered and nasobilliary drainage was obtained. After one week from the replacement of drainage and appropriate antibiotic and fluid replacement, a control ERCP was performed and no pancreatic leakage was detected. Consequently the drainage was moved out and the patient was discharged from the hospital after 4 days. The patient follow-up was done for two years in outpatient clinic bases and no complications were encountered till then.

Discussion

In adults, over 75% of blunt injuries to the pancreas are due to motor vehicle collisions[5].Blunt abdominal trauma may result in a variety of abdominal injuries most commonly involving the liver and spleen which are common and are usually detected by imaging without difficulty. Unfortunately pancreatic injuries may be more elusive and are uncommon. They may be overlooked in patients with extensive multi organ trauma and are associated with high morbidity and mortality due to fistula, abscess, sepsis, and hemorrhage, particularly if diagnosis is delayed [6, 7].

The pancreas is particularly vulnerable to crushing injury in blunt trauma due to impact against the adjacent vertebral column [8]. Two-thirds of pancreatic injuries occur in the pancreatic body, and the remainder occurs equally in the head, neck, and tail [9]. Yet in our case the trauma to the pancreas caused a serious fracture at the pancreatic body where the portal vein crosses the pancreas in addition with a spleen injury. The associated injuries as seen in our patient with spleen (could be liver, duodenum etc.) can occur in over 90% of the cases[3] .

Pancreatic injuries may be difficult to diagnose clinically because of the retroperitoneal location of the organ which mutes the clinical features of peritonitis.

Routine laboratory tests are of little help except for the elevation in serum amylase levels but they are neither specific nor sensitive for the diagnosis of pancreatic fracture [10]. In addition, the classical triad of fever, leukocytosis, and elevation of serum amylase levels is rarely encountered[11] . Elevation of serum amylase levels may be seen in only up to 73% of cases [3] . In our case the only laboratory findings were the anemia caused by internal bleeding and an elevated level of serum amylase which is found to be 800 iu/i. Actually, elevated levels of amylase focus our evaluation for further investigation of pancreatic injury.

Ultrasound scan is beneficial in emergency settings for the detection of free intra peritoneal fluid or a large hematoma that could consider a pancreas and associated vascular injuries, but

its capacity to show a specific damage to the pancreatic duct is limited[12].

In our case, the most helpful technique in the diagnosis of pancreatic fracture was depended on CT imaging and MRCP results. Advances in CT technology have enabled excellent demonstration of pancreatic parenchymal injuries. Helical multi-slice CT, which has both sensitivity and specificity as high as 80%, represents the best noninvasive diagnostic method for the detection of pancreatic injury. However, particularly in the initial phase, CT may miss or underestimate the severity of the damage because its accuracy in detecting major ductal injury is low. If the CT findings are suspicious or pancreatic injury possibility remains, MRCP may provide a clear demonstration of pancreatic duct disruption . In the past, ERCP was the only method available for evaluating pancreatic duct integrity[13]. The difficulty in performing ERCP in the acute setting, along with the associated risk of inducing iatrogenic pancreatitis in a traumatized patient, does not make this option favorable to most centers as an initial diagnostic tool.

The advantages of MRCP include noninvasiveness and greater availability than ERCP (endoscopic retrograde pancreatography). The main pancreatic duct can be identified by MR pancreatography within the pancreatic head in up to 97% of cases and within the pancreatic tail in up to 83% [14]. In addition, MRCP allows evaluation of the liver parenchyma and may demonstrate associated fluid collections. Moreover MR pancreatography can be helpful in directing ERCP-guided therapy when ductal anomalies are present, such as pancreas divisum which was present in our patient.

We have also benefit from MRCP and ERPC techniques in late postoperative complications of distal pancreatography where we were able to replace stent drainage to prevent pancreatic fluid leakage.

The degree of blunt pancreatic injury may be classified with the scheme developed by Moore et al.[15] as follows : grade A, pancreatitis or superficial laceration (50% pancreatic thickness); grade B1, deep laceration (50% pancreatic thickness) of the pancreatic tail ; grade B2, transection of the pancreatic tail; grade C1, deep laceration of the pancreatic head; and grade C2, transection of the pancreatic head. Guided by these classification guidelines, treatment may vary from simple drainage to performing Whipple's procedure. The grade of pancreatic injury consists of an independent predictor of both pancreatic complications and mortality [16]. We have performed a distal pancreatectomy in our patient with a diagnosis of a grade C2 injury. The incidence of distal pancreatectomy is reported to be 46% among pancreatic injury related cases in a series reported by Cogbill et al.[17]

Among cases treated surgically for pancreatic trauma, 20-40% will present complications [1]. In the short term, sepsis and multiple organ failure cause 30% of deaths after pancreatic trauma. After surgical treatment, secondary hemorrhage can originate from the pancreatic bed or the surrounding vessels as a result of retroperitoneal auto digestion. The formation of pancreatic fistulas is common. The incidence of abscess formation postoperatively as seen in our patient ranges from 10 to 25%. We have also encountered pancreatic fluid leakage even after re-laparotomy of the patient and fortunately were able to treat it with minimal invasive technique of ERCP guided stent placement before severe pancreatitis occurs. Mild pancreatitis may be anticipated in up to 18% of people who have undergone surgery for pancreatic trauma [17]. Endocrine and exocrine insufficiencies are very unusual after resection for pancreatic trauma. In the long term, pseudocyst formation can present weeks or months after the original injury.

Conclusion

Pancreatic injuries due to blunt trauma are uncommon but serious injuries that require early diagnosis to minimize complications and mortality. Therefore clinicians must be alert in multiple abdominal injuries and should always consider an associated pancreatic injury before or during laparotomy operations. . Any abdominal trauma patient suspicious of pancreatic injury due to USG or CT examination should promptly undergo MR cholangiopancreatography and/or ERCP should be performed for early diagnosis and prompt surgical intervention.

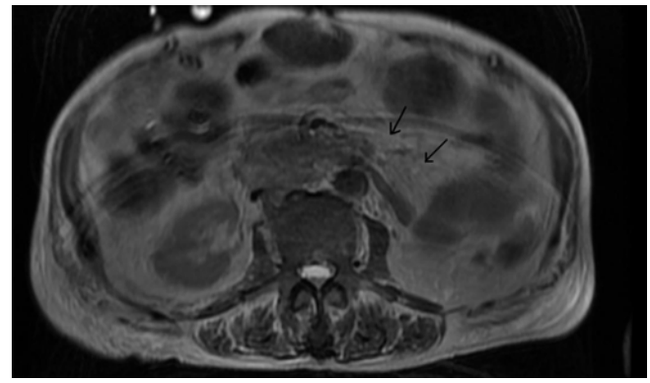

Figure 1. T2 weighted axial MR image shows full transection of the pancreas at the body with a fluid collection.

Competing interests

The authors declare that they have no competing interests.

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Case Report of a Left Atrial Myxoma Associated with Carney's Syndrome

Olgu Sunumu; Carney Sendromu İlişkili Sol Atriyal Miksoma

Carney's Syndrome and Left Atrial Myxoma

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Özet

Primer kardiyak neoplaziler, metastatik tümörlere oranla oldukça nadir görülürler. Primer kardiyak tümörlerin %70-%80'i benign miksomalardır. Miksomalar; kist ve mikroabse oluşumu, embolizasyon, senkop ve ani ölüm gibi komplikasyonlara yol açabilirler. Kardiyak miksomalar nadiren, primer nodüler adrenal kortikal hastalık, meme fibroadenomu, testis tümörleri, jigantizm ya da akromegali ile seyreden pituiter adenomaları ile birliktelik gösterebilirler. Bu duruma Carney sendromu adı verilir. Biz burada, Carney sendromu ile birliktelik gösteren ve sol atriyal miksoma tanısı ile ikinci kez ameliyat edilen bir vaka takdim etmekteyiz.

Anahtar Kelimeler

Carney Kompleksi; Miksoma; Pigmentasyon Bozuklukları

Abstract

Primary cardiac neoplasms are very rare as compared to metastatic tumors. 70% to 80% of them are benign myxomas. Complications of myxomas include cyst and microabscess formation, embolization, syncope and sudden death. Rarely, cardiac myxomas are associated with primary nodular adrenal cortical disease, mammary fibroadenomas, testicular tumors or pitiutary adenomas with gigantism or acromegaly known as Carney's syndrome. We present a patient with a left atrial myxoma who underwent reoperation associated with Carney's syndrome.

Keywords

Carney Complex; Myxoma; Pigmentation Disorders

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Introduction

Myxomas are the most frequent benign intracardiac tumors located mostly at the left atrium (75%) [1]. Myxomas mostly originate from the fossa ovalis. Other sites of origin are; the mitral annulus, mitral valve, aortic valve or the inferior vena cava [2]. It is known that myxomas can also cause embolisation, infection, syncope and suddent death [3]. Recurrence rates reported for cardiac myxomas are 4% to 7% for sporadic cases and 10% to 21% for familial cases [4]. In this case report, we present a patient who was operated for recurrent familial myxoma (Carney Syndrome).

Case Report

53 year old male patient was admitted with complaints of dyspnea and palpitations. He had a past medical history of hyperlipidemia, smoking and operation due to left atrial myxoma 15 years ago. His sister had been operated for myxoma aswell. There was a hyperpigmentation and nevus on the face and on the neck. On physical examination, there was a pansystolic murmur heard at the mesocardiac focus. The electrocardiography (ECG) was normal. The echocadiography revealed a left regular margined atrial mass (myxoma?) of 3.5x 2 cm diameter originating from the interatrial septum. A diagnosis of recurrent familial myxoma was made. Blood sample results for T3, T4, TSH, cortisol levels were within the normal ranges. Coronary angiography was performed to evaluate the patency of coronary artery disease. The patient had a %40 stenosis of the first diagonal branch of the left anterior descending artery. He was then referred to our cardiovascular surgery clinic for excision of the left atrial myxoma. The decision for operation was made for the left atrial myxoma associated with Carney's syndrome. The operation was performed under cardiopulmonary bypass, mild hypothermia using crsytalloid cardioplegia. A left atriotomy was done and the left atrial myxoma was visulalized (Figure 1). It originated from the interatrial septum. The myxoma was totally excised and the material was sent to the pathology department for microscopic investigation. The pathology report revaled a left atrial mass compatible with an atrial myxoma. The patient was discharged at the sixth postoperative day without any complication.

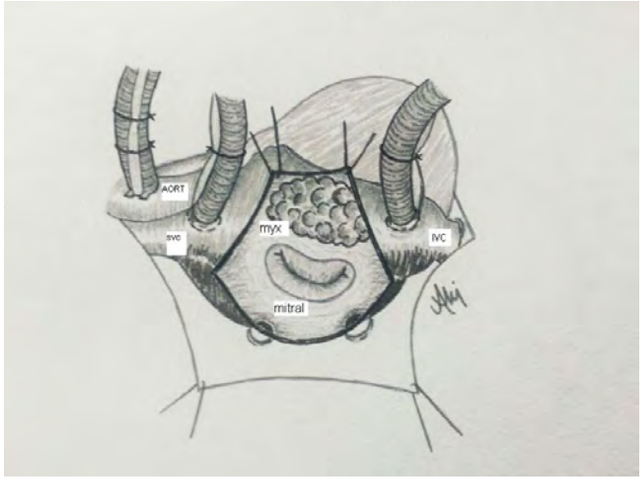

Figure 1. Illustration of intraoperative view of the left atrial myxoma.

Discussion

Myxomas are the most frequent primary heart tumors. The incidence of myxomas following autopsy series was reported as % 0.017 and % 0.028 (1). They are mostly located at the left atrium (%75). Surgical resection is the only way to treat myxomas and patients should be referred to surgery as soon as possible. Myxomas can present at any age group but occur more often between the 3rd and the 6th decades of life [5] as is the case with our patient who was 53 years. Familial forms, which are more frequently diagnosed in younger individuals, constitute 10% of all myxomas and have autosomal dominant transmission [6]. Familial cardiac myxomas are characteristically seen in atypical locations and have a high recurrence rate. First-degree family members should be screened and followed carefully. Recurrence rates reported for cardiac myxomas are 4% to 7% for sporadic cases and 10% to 21% for familial cases [4]. Carney's complex, as described by J.A. Carney, is characterized by the association of cutaneous pigmentation, fibromyxoid tumors of the skin, myxomas of the heart, endocrine overactivity, and autosomal dominant inheritance [5,7].

Our patient presented here had a hyperpigmentation and nevus on the face and on the neck. By the guidance of the physical examination findings and the familial history of the left atrial myxoma, the patient was diagnosed as Carney's sydrome and was treated succesfully by surgery.

Competing interests

The authors declare that they have no competing interests.

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Acute Generalized Exanthematous Pustulosis (AGEP) Induced by Cetirizine in a Child A Case Report

Çocukta Setrizin ile İndüklenen Akut Generalize Ekzantematöz Püstüloz

AGEP Induced Cetirizine in Childhood

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Özet

Akut Generalize Ekzantematöz Püstüloz (AGEP), yaygın foliküler olmayan steril püstüller ile karakterize nadir bir kutanöz döküntüdür. AGEP çocuklarda az rastlanılan bir hastalıktır ve genellikle ilaçlara bağlı ortaya çıkmaktadır. Antibiyotikler, sulfonamidler, ateş düşürücü ve ağrı kesiciler bu ilaç döküntüsünün en sık nedenleridir. Setrizin çocuklarda angioödem, atopik dermatit ve ürtiker tedavisinde sıklıkla kullanılan ikinci kuşak antihistaminiktir. Literatürde ise setrizin ile indüklenen AGEP olgusu yer almamaktadır. Bu olguda oniki yaşında kız çocuk gövdede yer alan ürtikeryal plaklar ile başvurmuş ve tedavi olarak kullanılan setrizin (günde tek doz) ile foliküler olmayan püstül formasyonu gelişmiştir. Tedavinin sonlandırılmasından sonlandırılmasının ardından deskuamasyon ile püstüller tamamen gerilemiş ve oral provakasyon testine yanıtta pozitif olarak alınmıştır. Bu olguda çocukluk yaş grubunda antihistaminiklerin (setrizin) AGEP nedeni olabileceğini göstermek amacıyla sunulmuştur.

Anahtar Kelimeler

AGEP; Çocuklukçağı İlaç Reaksiyonları; Setrizin

Abstract

Acute Generalized Exanthematous Pustulosis (AGEP), is a rare cutaneous rash characterized by widespread sterile non-follicular pustules. AGEP is a rare disease in childhood and it is often due to drugs. Antibiotics, sulphanamides and antipyretic-analgesics are the main reasons of this drug reaction . Cetirizine is a second generation antihistamine is often used in the treatment of angioedema, atopic dermatitis and urticaria in children. Cetirizine induced AGEP was not reported in the literature. In this case a twelve year old child was admitted with urticarial plaques located on her trunk. She developed maculopapular lesions and pustular eruption with Cetirizine (once a day) treatment. Cetirizine was stopped and the nonfollicular pustules cleared with a desquamation. The result of the oral challenge test was positive. We present this rare case to show that the antihistamines (cetirizine) may cause AGEP in childhood.

Keywords

AGEP; Cetirizine; Childhood Drug Reactions

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Introduction

Acute generalized exanthematous pustulosis (AGEP) is a rare acute reaction that is drug induced in 90% of cases.[3] It is characterized by widespread, sterile pustular rash. The eruption is of sudden onset and appears 7-10 days after the medication is started. Pustules resolve spontaneously within a few days by cessation of the drug.[4] Cetirizine is a second generation antihistamine which is used to treat urticaria, atopic dermatitis and angioedema in childhood.[2] This case is important to indicate the cetirizine as a causative factor of AGEP in children.

Case Report

A twelve year old girl was admitted with erythematous oedematous plaques on the chest and extremities and complaining of pruritus. She had no drug history. The patient was taken to the hospital and treated with methylprednisolone (1 mg/kg/day) and cetirizine (daily) also she did not take any other medication. Her urticarial lesions resolved on the fourth day of the treatment. Although the medication had not been changed, on the sixth day she presented with a pruritic, erythematous maculopapular eruption affecting the abdomen, neck and intertriginous areas. One day later she developed pustular lesions and her temperature was 37 C^0. Neutrophils were 12.500 cells/æl (range 1.700-8.000 cells/æl). Histopathologic examination showed subcorneal pustules with inflammatory infiltrate consisting of neutrophils in epidermis and superficial dermis. (Figure 1-2) Methylprednisolone was continued and cetirizine was stopped. After withdrawal of cetirizine and introduction of desloratadine, the disseminated nonfollicular pustules cleared within three days following a desquamation. Oral challenge was done with cetirizine and the pustuler eruption occurred again with the same distribution. The patient was asked about previous adverse reaction to other drugs and no personal or family history of drug reactions and no history of psoriasis was evident.

Discussion

AGEP is a disease characterized by the rapid onset of many sterile, nonfollicular pustules, often erythema, develops very acutely and erythema soon dozens to hundreds of small nonfollicular sterile pustules predominantly located on face, trunk and lower limbs.[1]

Skin symptoms are almost accompanied by fewer above 37 C^0. Leucocytosis is mostly due to blood neutrophil counts above 7x109 /l. After the withdrawal of the drug, pustules resolve spontaneously within a few (4-10 days) days and are in typical cases followed by a characteristic post-pustular pin-point desquamation.[1,3,4]

It seems that more than 90 % of cases with AGEP are drug induced. A wide range of drugs has been suspected of causing these reactions and antibacterials are being the most frequent triggers.[1] The disease is usually caused by penicillins or macrolides. In a minority of cases viral infections have been suspected to trigger AGEP.[5] Typical histopathology shows spongioform subcorneal and/or intraepidermal pustules, and often marked perivascular infiltrates with neutropils.[3] Also AGEP is a disease that is rarely seen in childhood.[3] In this case the onset of the disease about five day after the administration of the cetirizine, neutrophylia, typical histopathologic examination and rapidly response to cessation of the drug lead us to the diagnosis of AGEP. The main differential diagnosis clinically and histologically is pustular psoriasis. In a number of reports patients had a history of psoriasis, however many studies agree with that AGEP is not associated with psoriasis.[4]

Antihistamines are the rare causes of the drug reactions and cetirizine induced AGEP was not reported in the literature. This rare case report is so important that clinicians should keep the possibility of this cutaneous drug reaction in mind while choosing antihistamines for children.

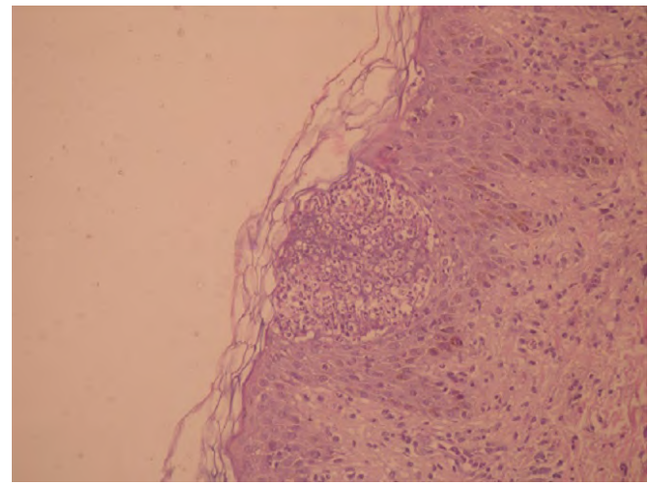

Figure 1. Subcorneal pustule formation in biopsy

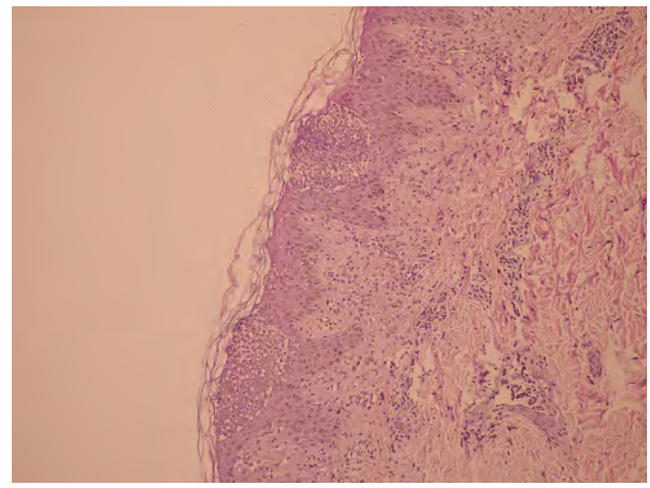

Figure 2. Perivascular neutrophylic infiltration

Competing interests

The authors declare that they have no competing interests.

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Case Report
JCAM

Low Dose Methotrexate Induced Perilesional Bullous Erythema in Plaque Type Psoriasis

Plak Tip Psöriazisde Düşük Doz Metotreksat ile İndüklenen Perilezyonel Büllöz Eritem

Perilesional Bullous Erythema

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Bu çalışma 9th EADV Spring Symposium, June 6-10, 2011, Verona, İtalya'da poster olarak sunulmuştur.

Özet

Kemoterapi ile indüklenen büllöz tip akral eritem, metotreksat ile ve daha sıklıkla sitozin arabinozid ile bildirilmiştir. Ancak metotreksat ile ilişkili perilezyonel büllöz eritem daha önce bildirilmemiştir. Burada, biyopsi ile kanıtlanmış jeneralize plak tip psöriazisi olan 64 yaşında bir erkek hastada 15 mg/hafta dozda metotreksat tedavisi sonrası büllöz perilezyonel eritem gelişimi sunuldu. Hastada tedavinin ilk 3 günü içerisinde psöriaziform plakları çevreleyen simetrik, iyi sınırlı, ağrılı, eritemli perilezyonel büllöz lezyonlar gelişti. Lezyonlar güçlü topikal kortikosteroidlere ve ıslak pansumanlara yanıtsız idi. Üç hafta sonra topikal kortikosteroidler kesildi ve metotreksat dozu 10 mg/haftaya azaltıldı. Büllöz perilezyonel eritemin devamı nedeniyle, anti-inflamatuar yanıt için kına ekstresi "Lawsonia inermis" içeren bir topikal bitkisel tedavi başlandı. Lezyonlar giderek iyileşti ve iki hafta içerisinde rezidüel hiperpigmentasyon ile tama yakın geriledi. Metotreksat ile tedavi olan psöriatik olgularda, nadir olarak perilezyonel büllöz eritemin görülebileceği ve metotreksat tedavisinin kesilmesinin gerek olmadığını düşünmekteyiz.

Anahtar Kelimeler

Büllöz Lezyon; Eritem; Lawsonia Bitkisi; Metotreksat; Psöriazis

Abstract

The bullous variant of chemotherapy-induced acral erythema has been reported with methotrexate and more frequently cytosine arabinoside. However, perilesional bullous erythema in association with methotrexate hasn't been reported before. Herein, we presented a 64-year-old male patient, a biopsy proven case of generalized plaque psoriasis, who developed a bullous perilesional erythema after a single oral dose of 15 mg/week methotrexate. The patient developed symmetrical, well-demarcated, painful, erythematous perilesional bullous lesions surrounding these psoriasiform plaques within 3 days of receiving the medication. The lesions were unresponsive to the potent topical corticosteroids and wet dressings. After 3 weeks the topical corticosteroids were discontinued and methotrexate dose was reduced to 10 mg/week. As a result of the permanency of the bullous perilesional erythema, a topical herbal therapy including a henna extract "Lawsonia inermis" was started for an anti-inflammatory response. The lesions gradually improved and resolved almost completely with residual hyperpigmentation within two weeks. We believe that, perilesional bullous erythema may be seen rarely in psoriatic patients treated with methotrexate and there is no need for discontinuation of methotrexate therapy.

Keywords

Bullous Lesion; Erythema; Lawsonia Plant; Methotrexate; Psoriasis

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Introduction

Chemotherapy-induced acral erythema (CIAE) is characterized with a painful erythema of the palms and soles which occurs following chemotherapy due to cytarabine, paclitaxel, mercaptopurine, doxorubicin and fluorouracil [1]. The bullous variant had been reported in relation to methotrexate (MTX) and more frequently cytosine arabinoside treatment [1-3]. The reaction usually begins with palmoplantar dysesthesia, causing symmetrical erythema with well-demarcated borders and blistering and eventual desquamation that remains limited to the palms and soles [1,4]. To the best of our knowledge, up to now almost fifteen case reports of bullous acral erythema (AE) due to MTX have been published, but this is the first case of MTX induced perilesional bullous erythema [2,3,5-7].

Case Report

A 64-year-old man with a biopsy proven case of plaque type psoriasis for 15 years was unresponsive to topical treatments of corticosteroids, calcipotriol and PUVA. Therefore oral methotrexate 15 mg/week, with 5 mg of folinic acid per day was prescribed after the routine blood examinations. His other medical and drug history were unremarkable. On the dermatological examination; several demarcated, erythematous psoriasiform squamous plaques were present on both shins and elbows. No other chemotherapeutic or immunosuppressive agents were additionally administered. The patient developed symmetrical, well-demarcated, painful, erythematous perilesional bullous lesions surrounding these psoriasiform plaques within 3 days of receiving the medication (Figure 1). His palms and soles were

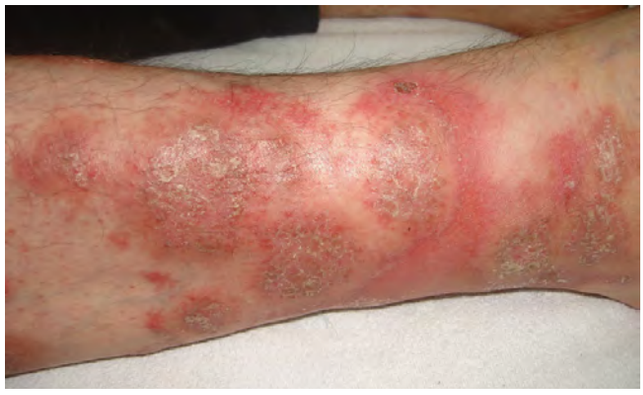

Figure 1. Well-demarcated, painful, erythematous perilesional bullous lesions surrounding these psoriasiform plaques within 3 days of receiving the medication.

spared and no mucosal lesions were noted. His blood count, liver and kidney function tests and urine analysis were all normal. No systemic side effects were observed.

The histopathological examination of the punch biopsy material taken from the bullous lesion showed acantholysis, subepidermal bullae with keratinocyte necrosis, apoptosis, vacuolar changes along the basal cell layer and lymphocytic inflammatory infiltration in the upper dermis. Direct immunofluorescent staining was negative for C3 and Ig G. Serum C3 and Ig G antibodies detected by indirect immunofluorescent study were also negative. He was diagnosed with MTX induced perilesional bullous erythema. Written informed consent was obtained from the patient for publication of this brief case report and accompanying images.

The patient was first treated with potent topical corticosteroids and wet dressings without any response for 3 weeks intermittently. After one month the topical corticosteroids were discontinued and methotrexate dose was reduced to 10 mg/week and a topical henna extract (Kapederm® cream) 6 times a day was started for an anti-inflammatory response. The lesions gradually improved and resolved almost completely with residual hyperpigmentation within two weeks (Figure 2). No cessation of

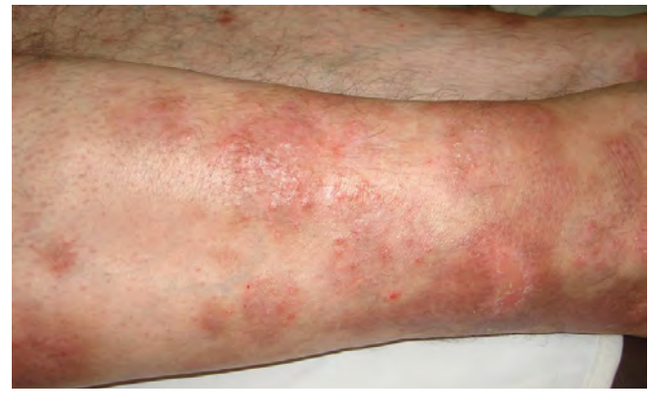

Figure 2. The lesions gradually improved and almost resolved completely within two weeks with topical herbal therapy including a henna extract "Lawsonia inermis".

the treatment was required. His pain completely resolved and no recurrence was noted in 10 months follow up.

Discussion

On the contrary that CIAE is also called as a "palmoplantar erythrodysesthesia syndrome" or "chemotherapy-induced syringosquamous metaplasia" caused by the toxicity of the chemotherapeutic agents concentrated in the eccrine sweat glands, the sparing of the palmoplantar areas was very unusual in this case [4]. This reminds us another possible etiopathogenic mechanism like the presence of an associated local inflammatory receptor in the psoriatic plaque lesions that links the systemic toxic agent causing a bullous reaction. But on this occasion, we should have expected more severe cases of chemotherapy induced bullous erythema after the intralesional injection of MTX in plaque type psoriasis. We believe that the present new entity may be re-named as not CIAE but "chemotherapy induced perilesional bullous erythema in an erythema multiforme like pattern". However, furthermore similar cases are also needed to be investigated. In this case we also excluded the diagnosis of autoimmune bullous diseases, as a result of the lack of the histopathological findings and negativity of the direct and indirect immunofluorescent tests.

In the prior reported cases of MTX induced bullous AE including this case the lesions appeared rapidly within the first 24-72 hours [2]. It is usually dose dependent and mostly appears with bolus high-dose infusions or long-term low-dose infusions [4]. As we know, CIAE due to oral, low dose (15 mg/week) and short-term MTX treatment was very unusual.

The patients with CIAE usually recover without complications [1,4]. Response to dose reduction is expected but not to folinic acid. Interestingly, our patient was almost completely ameliorated with topical use of henna in two weeks without cessation of the MTX therapy. "Henna" is a dye extracted from the dried leaves and skin of branches of a bush-like plant from lythracea

family called “Lawsonia inermis” which is used in eastern cultures and in rural areas of Turkey. So far anti-microbial, anti-inflammatory, anti-oxidant and immunomodulatory effects of henna have been reported in rats [8].

In a previous case series including 10 patients with hand and foot syndrome (HFS) due to capacitabine chemotherapy remarkable and rapid clinical improvements were also reported after using topical henna like in our case [9]. Of those patients with Grade 3 HFS, in four; complete response, in two; regression to grade 1 level were reported. The complaints of the other four patients with grade 2 HFS were also fully recovered after the first week of outpatient visit [9]. In an animal model, it was also demonstrated that “Lawsonia inermis” extracts are capable of promoting wound healing activity [10]. Enhanced wound contraction, tensile strength, increased hydroxyproline content in histological observations suggest that “Lawsonia inermis” has potential in the management of wound healing and these invite further studies. The sudden dramatic improvement of this patient with topical henna suggests us that it might act as an anti-inflammatory agent.

In conclusion, the clinical improvement in this case might be a spontaneous resolution seen in CIAE reactions, or it might be due to elimination of another unknown causative agent. The reduction of the MTX dose from 15 to 10 mg/week might also play a role in improvement of the lesions in this case. We also approved to emphasize that the improvement of the lesions suggests that topical henna might act as an anti-inflammatory agent.

We believe that, perilesional bullous erythema may be seen rarely in psoriatic patients treated with MTX and there is no need for discontinuation of MTX therapy.

Competing interests

The authors declare that they have no competing interests.

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Missed Abdominal Wall Abscess in a Child. Neglected Clinical Examination and Improper use of Ultrasonography?

Çocukta Karın Duvarı Absesi. Klinik Muayene ve Ultrasonografi Uygunsuz Kullanımı?

Abdominal Wall Abscess

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Özet

Akut karın ağrısı çocuklarda birçok tanı ve yönetim sorunları oluşturmaktadır. Tam bir sonografik klink tanı için klinisyen tarafından ayrıntılı klinik muayene ve uygun rehberlere ihtiyaç vardır. Biz akut karın ağrısına neden olan tanısı yanlış konulmuş karın duvarı apseli, klinik muayene ve uygun rehber kullanılmamış bir çocuk olguyu sunduk.

Anahtar Kelimeler

Karın Ağrısı; Klinik Muayene; Tanı; Ultrasonografi

Abstract

Acute abdominal pain pose many diagnostic and management problems in children. There is need for a thorough clinical examination and proper guidance by clinician to the sonologist for the diagnosis. We report a child with acute abdominal pain due to abdominal wall abscess misdiagnosed because of lack of clinical acumen and improper use of ultrasonography.

Keywords

Abdominal Pain; Clinical Examination; Diagnosis; Ultrasonography

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Introduction

Abdominal pain is a common problem in children and pose a diagnostic dilemma(1). Ultrasonography has become a key diagnostic tool in acute pediatric abdominal pain(2). Eventhough many investigations have been introduced, clinical decision-making is very important. We report a 9 year old child with acute abdominal pain due to abdominal wall abscess misdiagnosed because of lack of clinical accumen and improper use of ultrasonography without clinicians guidance.

Case Report

9 year old female presented with abdominal pain since 5 days, fever and non bilious vomiting since 1 day. Abdominal pain was epigastric, intermittent, non radiating and not related to food. There was no bowel or bladder disturbances. Before presenting to us she was seen by surgeon/pediatric surgeon and diagnosed as acute gastritis/cholecystitis and two sonologists opined as normal abdominal sonography. On examination she was febrile(1010F), had no jaundice and was hydrated well. Abdominal examination revealed firm, tender, discrete swelling (1*1. 5cm)in the epigastrium just below the xyphisternum extending towards the right hypochondrium. Coughing/ movement of right chest wall aggravated the pain. Leg raising test(making the abdominal muscles tense) demonstrated that the swelling is not intraabdominal and Carnett's test was positive(increased tenderness). Other systemic examination was unremarkable. Investigations:-Hb11. 3g/dL, TLC 17400/mm3 (Neutrophils 83%, Lymphocytes17%), platelets 4. 08lakhs/mm3, sugar 110mg/dL, HIV negative, Mantoux test negative and normal chest X-ray. Ultrasound done within 24 hours by two different sonologists was opined as normalabdominal sonography. Again within 12 hours resident accompanied the child for sonography from a third sonologist. This time sonologist detected heterogenous lesion(abscess) measuring 3. 4*2cm noted with few hypoechoic areas just anterior to right rectus muscle in the epigastric region[Fig. 1]. Probably the sonologist would have missed the finding but for the pediatric resident who inform the sonologist of his clinical impression, guided regarding the correct placement of the probe and sonologist also agreed to that. Child was started on Inj Co-amoxiclav and ceftriaxone. Incission and drainage of pus was done and she was discharged after 5 days.

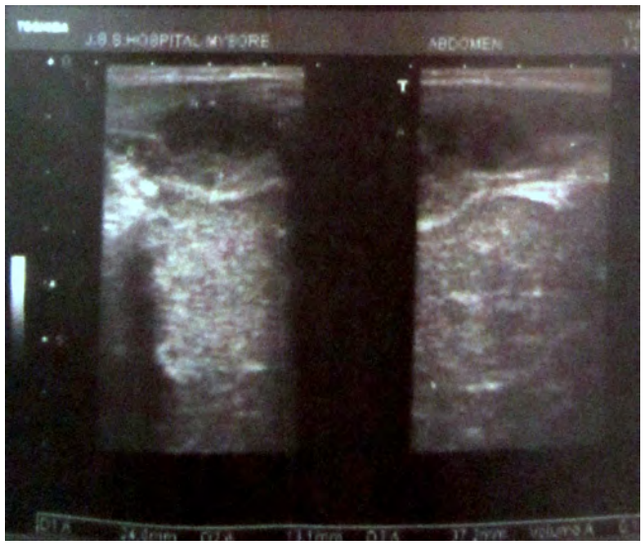

Fig 1. Abscess measuring 3. 4*2cm noted with few hypoechoic areas just anterior to right rectus muscle in the epigastric region.

Discussion

Acute abdominal pain is a common problem in children and it may herald a surgical or medical emergency(1). The most difficult challenge is making a timely diagnosis so that treatment can be initiated and morbidity prevented. The abdominal wall as a source of pain has received little attention and overlooking can result in a expensive and dangerous error in evaluation(3). Abdominal pain clinically falls into three types: intraabdominal pain(visceral), abdominal wall pain(parietal) and referred pain. Visceral pain usually is dull, poorly localized, and felt in the midline. Parietal pain usually is sharp, intense, discrete, localized and coughing or movement can aggravate it(1). In fact our child had classical findings of parietal pain. , but because it was epigastic it lead to the erroneous diagnosis. Primary infection of the abdominal wall is rare and can have symptoms mimicking those of an acute abdomen(4). Abdominal wall lesions frequently present as palpable masses and often mimic intra-abdominal conditions(5). Physical findings in the abdominal wall pathologies have low specificity and often a clinically suspected intra-abdominal lump proves to be in the abdominal wall(5). Clinical examination plays a key role in determining which children should undergo immediate surgical consultation for appendicectomy and who should undergo further diagnostic evaluation, including diagnostic imaging(6). However, the importance of clinical examination have become submerged beneath lot of investigations now a days. Carnett's test is a useful clinical test for differentiating abdominal wall pain from intra-abdominal pain(3). If tenderness is unchanged or increased when abdominal muscles are tensed [by straight-leg-raising or head raising maneuver in the supine position while the examiner's hand touches the painful site] (positive Carnett's sign), the abdominal wall is the likely origin of pain. In contrast true visceral sources of pain are associated with less tenderness. Carnett's test has a sensitivity of 81% and a specificity of 88%. This suggests that clinical methods are sufficient to diagnose abdominal wall pain in most cases and carnett's test saves numerous unnecessary and often unpleasant investigations(7). Out of 120 acute abdominal pain cases, 23 of 24 patients with a positive Carnett's sign had a normal laparotomy(8). In our case also diagnosis was missed because of not eliciting the carnett sign and the presence of a tiny mass near xyphisterum. Usually when a child presents with fever, vomiting and pain abdomen clinician think mainly acute appendicitis & cholecystitis and tries to look for tenderness in the umbilicus /right iliac fossa/right hypochondrium not near xyphisternum. In our case fever is mainly due to abdominal wall abscess and probably because of epigastric tendeness, acute gastritis & cholecystitis diagnosis was made initially. Ultrasound is usually the initial diagnostic imaging modality in cases of palpable abdominal masses to determine their nature, and their localization, either intra-abdominally or in the abdominal wal(9). Computed tomography or MRI may be used for additional characterization(9). Ultrasound can establish its presence, primary origin, size, extent and effect on the surrounding structures. Ultrasound is simple, quick, painless, noninvasive, without ionising radiation and side effects(10).

The outcome of ultrasound findings has a significant bearing on the management of the patient and choice of subsequent radiological investigations, if needed(10). With the introduction of high-frequency, high-resolution probes. , recognition of different layers of the abdominal wall is now possible on USG examinations(5). However, since the abdominal wall is a superficial structure, a mass within it can be easily overlooked if a water-path is not used to increase the distance between the transducer surface and the skin(10). Ultrasound examinations were done to evaluate clinically palpable abdominal masses in 125 Children and in 15 patients, the clinically palpable masses were actually anterior abdominal wall abscesses or hernias(10). However, since ultrasonography is operator dependent, the expertise of the sonologist would also be contributory to the outcome(10). Before subjecting the case for sonography clinician should guide the sonologist about the differential diagnosis so that sonologist can select the appropriate probe/transducer/machine. Unless specified about the abdominal wall pathology sonologist may not use high frequeny sonography and miss the abdominal wall lesion, same situation occured in our case also. First two sonologists missed the diagnosis by not using high frequeny machines and appropriate transducers. Third sonologist could diagnose, as the resident who accompanied the child could throw the light on the clinical findings and even the third sonologist accepted this fact. To conclude acute abdomen can be managed ethically in a fruitful way by good clinical examination, guiding the sonologist about the differential diagnosis and what to look for in a given patient thereby avoiding unnecessary, dangerous invasive investigations and laparotomies.

Competing interests

The authors declare that they have no competing interests.

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Inflammatory Myofibroblastic Tumor of the Prostate

Prostatın İnflamatuar Myofibroblastik Tümörü

Inflammatory Myofibroblastic Tumor

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Özet

Prostatın inflamatuar myofibroblastik tümörleri (İMT), sarkomlar ve iğsi hücreli karsinomları klinik ve histopatolojik olarak taklit edebilen nadir lezyonlardır. Burada sunulan olgu, normal prostat spesifik antijen düzeyleri ve kronik yakınmaları olan, tıbbi tedaviye yanıtsız infravezikal tıkanma bulguları nedeni ile suprapubik prostatektomi uygulanan 63 yaşında bir hastadır. Eksizyon materyalinin histopatolojik incelenmesi fokal nükleer pleomorfizm, hiperkromazi gösteren, mononükleer iltihabi infiltrasyon ve miksoid değişiklikler içeren, düzgün sınırlı, iğsi hücreli bir lezyonu ortaya koydu. Mitoz nadirdi. İmmunhistokimyasal çalışmada düz kas aktini ve vimentin pozitif, anaplastik lenfoma kinaz -1 fokal pozitif, S-100 ve pansitokeratin negatifti. Lezyon inflamatuar myofibroblastik tumor olarak tanı aldı. İMT'nin prostatın malign iğsi hücreli lezyonlarından ayrımı gereksiz ileri tedavi işlemlerinin önüne geçilmesi için şarttır.

Anahtar Kelimeler

İnflamatuar Myofibroblastik Tümör; Prostat; Psödosarkomatöz; Fibromiksoid; Psödotümör

Abstract

Inflammatory myofibroblastic tumors (IMT) of the prostate are very rare lesions that may mimic sarcomas and spindle carcinomas both clinically and histopathologically. The case presented here is a 63-year-old patient, with normal prostate specific antigen levels and a chronic history of complaints, who underwent to suprapubic prostatectomy due to the infravesical obstruction symptoms that are resistant to medical therapy. Histopathological examination of the excision material revealed a well demarcated spindle cell lesion with focal nuclear polymorphism, hyperchromasia, mononuclear inflammatory infiltration and myxoid areas. Mitosis was rare. Immunohistochemically smooth muscle actin and vimentin were positive, anaplastic lymphoma kinase-1 was focal positive, S-100 and pancytokeratin were negative. The lesion was diagnosed as an inflammatory pseudotumor. Differential diagnosis of the IMT from malignant spindle cell tumors of the prostate is crucial to prevent overtreatment.

Keywords

Inflammatory Myofibroblastic Tumor; Prostate; Pseudosarcomatous; Fibromyxoid; Pseudotumor

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Introduction

Stromal lesions of the prostate are rare and consisted of a number of benign and malignant lesions that closely resemble each other [1,2]. The differential diagnosis, which is crucial for appropriate treatment, may be challenging. Inflammatory myofibroblastic tumor of the prostate (IMT; synonym: pseudosarcomatous spectrum fibromyxoid tumor of the prostate, inflammatory pseudotumor of the prostate) is one of these rare lesions and stands in the benign side of the stromal lesions.

Case Report

A 63-year-old male patient has admitted to the department of urology with difficulty in starting urine flow and pollakiuria for 2 years. Despite the 6 months of 5 alpha reductase (Finasterid 5mg) and alpha blocker (Tamsulosin HCL 0.4mg) medication, the symptoms persisted. Prostate specific antigen (PSA) was 1.22 ng/ml, and prostate volume was measured as 186cc in ultrasound. In uretherocystoscopia prostatic urethra was longer than normal (7cm) and prostate median lobe and lateral lobes were narrowing the urethra. Qmax, volume and residue in uroflow, performed without Foley catheter, were 12ml/sec, 75cc and 197 cc respectively. Bladder capacity, compliance, and sensitivity were normal in filling cystometry. There was no detrusor hyperactivity. Pressure flow study showed high pressure-low flow. Digital rectal examination finding was compatible with a 1.5(+) adenoma.

The patient had no history of a previous operation or instrumentation.

Since the patient was not an appropriate candidate for transurethral resection, suprapubic prostatectomy was performed with a clinical presumptive diagnosis of benign prostatic hyperplasia.

Macroscopically the resection material was composed of two separate tissues (originally a single mass resected in two portions by the surgeon because of its size) with smooth surface, gray-white in color. One of the pieces was 9x4x4cm and the other was 7x5x4cm, total weight: 141gr. The cut surfaces were solid, gray-white in color and had a fibrillary appearance. Extensive sampling was done.

Microscopically the well demarcated lesion that was composed of spindle cells forming long, transecting bundles, was pushing, but not infiltrating the normal prostate tissue. Focal nuclear hyperchromasia and pleomorphism were observed. Mitosis was rare. There was no necrosis or atypical mitotic figures. There were foci of mononuclear inflammatory response, including plasma cells, and myxoid degeneration (Figure 1). Vimentin, smooth muscle actin were positive, anaplastic lymphoma kinase -1 (ALK-1) was focal positive, S-100 and pancytokeratin were negative in immunohistochemical study. ALK positivity was confirmed with fleurocein in-situ hybridization (FISH) with DAKO split signal FISH probe (Figure 2).

The case was diagnosed as inflammatory myofibroblastic tumor. The patient is under follow up, for 5 years and no recurrence occurred.

Discussion

Inflammatory myofibroblastic tumors are rare lesions that can be seen in a variety of organs (e.g. lungs, heart, liver, vagina, kidneys) [2, 3]. The ethiopathogenesis is uncertain. The term "inflammatory pseudotumor" was first used by Umiker and Ivenson in 1954 for desricribing 4 cases in lung [2]. Compared with the respiratory system the term "inflammatory pseudotumor" is relatively new in genitourinary tract. First "reactive pseudosarcomatous response" in genitourinary tract was described in bladder in 1980 [4], and the first IMT in prostate was described 4 years later by Hafiz et al [5].

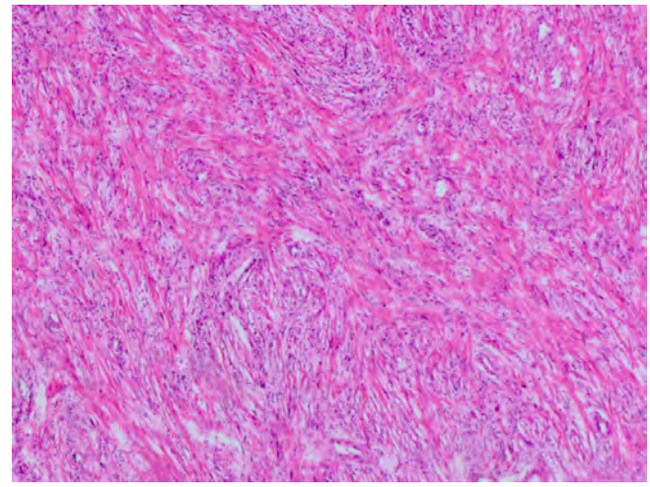

Figure 1. Photomicrograph reveals transecting spindle cell bundles intermixed with mononuclear inflammatory infiltrate (HE x100)

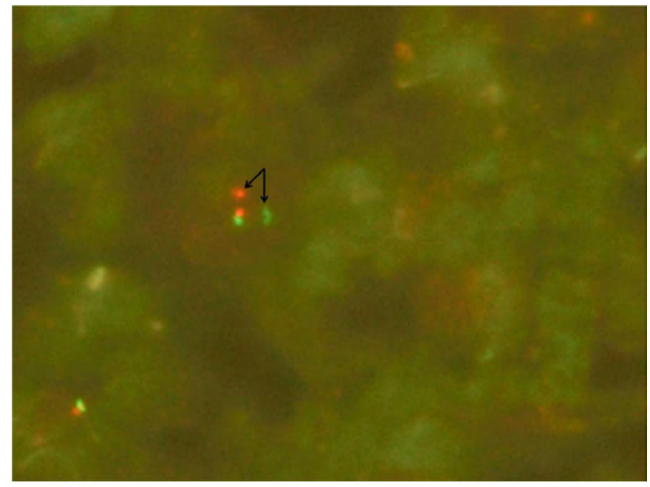

Figure 2. Signal splitting in FISH. Note one green and one red signal splitted from each other by a distance more than two signal diameters (arrows) (ALK FISH x1000)

IMTs are spindle cell lesions that are believed to be originate from the myofibroblastic cells [6]. They may mimic malignant lesions both clinically and microscopically. In clinic, IMTs may show rapid grow and high PSA levels. Microscopically they may be confused with sarcomas and spindled carcinomas, which will lead to cystoprostatectomy and pelvic lymph node dissection. In our case the PSA levels were not suggestive of malignancy and the clinical symptoms of the patient were existing nearly for two years, without a rapid increase. The size of our lesion is larger than the previously reported IMTs of prostate [1, 7]. This slow pace of growth may explain the relatively larger size of the lesion. If the lesion showed rapid growth and caused more dramatic symptoms, the patient would have admitted to the hospital earlier and probably the lesion would be excised in an earlier phase. In microscopic examination there was focal nuclear pleomorphism and hyperchromasia. Mitosis was rare

and necrosis was absent. In sarcomas and spindled carcinomas, nuclear pleomorphism and hyperchromasia are more prominent with a high mitotic rate and invasive pattern. Foci of necrosis may also be present.

Postoperative spindle cell nodule (PSCN), and IMT were thought to be different entities, but now they are believed to be reflecting the same lesion with some minor differences, particularly in patient history. PSCN develops after an instrumentation or operation, and tends to be smaller than IMT [1, 6]. Our patient had no history of a previous operation or instrumentation.

Embryonal, fibroblastic, and smooth muscle nodules are the stromal nodules of benign prostatic hyperplasia (BPH) [1]. They are all well demarcated nodules and may contain pleomorphic spindle cells and hyperchromatic pleomorphic nuclei [8], like IMT. All BPH nodules are positive for S-100 [1]. In our case S-100 was negative. Although one may speculate that it may be possible that some IMT nodules were being reported as BPH nodules in the past, and the real incidence of IMT may be higher than the current data, after nearly 30 years of its first description, there is still a small number of case of reports in the literature regarding prostatic IMTs.

Immunohistochemical ALK-1 positivity may be seen in genitourinary tract, most commonly in bladder, IMTs. In a study by Montgomery et al, FISH ALK results and immunohistochemical ALK results were showed to be compatible [7]. The ALK positivity suggests that IMTs of genitourinary tract are true neoplastic processes rather than reactive ones. ALK positivity may not be a predictor of recurrence [7].

IMTs of prostate are rare lesions and their distinction from sarcomas and spindle cell carcinomas is crucial to prevent an overtreatment. Most IMTs of prostate are treated with transurethral resection, an interventional therapy that aims to resolve the symptoms rather than a total resection of the mass. Since IMTs are true neoplastic processes, close follow up of the patients with IMT diagnosis may be important.

Competing interests

The authors declare that they have no competing interests.

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Case Report
JCAM

Prenatal Diagnosis of Harlequin Ichthyosis: Report of a Case

Harlequın İktiyozisinin Prenatal Tanısı: Bir Olgu Sunumu

Harlequin Ichthyosis

Cem Yaşar Sanhal, Murat Özekinci, Mehmet Sakıncı, Mehmet Şimşek, İnanç Mendilcioğlu
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Bu olgu sunumu 11. Ulusal Jinekoloji ve Obstetrik Kongresinde (Mayıs 2013, Antalya) e-poster olarak sunulmaya hak kazanmıştır.

Özet

Sıklıkla neonatal dönemde ölümle sonuçlanan ve keratinizasyon bozukluklarının en ciddi formu olarak kabul edilen Harlequin iktiyozisi (Hİ), cildin keratin tabakasında derin yarıklar, vücudu zırh gibi kaplayan yoğun bir kabuk ve gözü, kulağı ve ağızı etkileyen kontraksiyon anormallikleriyle karakterizedir. Bu olgu sunumunda, akraba olan bir Türk çiftte tekrarlayan fetal Hİ ve prenatal ultrasonografik tanısı sunulmaktadır.

Anahtar Kelimeler

İktiyozis; Prenatal Tanı; Prenatal Ultrasonografi; Akraba Evliliği

Abstract

Harlequin ichthyosis (HI) - the most severe form of keratinizing disorders, often lethal in the neonatal period - is characterized by a profound thickening of the keratin skin layer, a dense armor-like scale that covers the body, and contraction abnormalities of the eyes, ears, and mouth. Here, we report a recurrent case of fetal HI and its prenatal ultrasonographic diagnosis in a Turkish consanguine couple.

Keywords

Ichtyosis; Prenatal Diagnosis; Prenatal Ultrasonography; Consanguinity

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Introduction

Harlequin ichthyosis (HI) is a severe disorder of keratinization caused by mutations in the ABCA12 gene with autosomal recessive inheritance [1]. The mutations lead to defective lipid transportation which negatively affects the correct development and function of the skin [2]. The characteristic clinical features of HI include thick, plate - like scales over the entire body with ectropion, eclabium and flattened ears [3]. The overall survival rate of this severe disorder is 56% [4]. Retinoids (etratinate, acitretin) are the major drugs that are generally preferred in postnatal management of HI [5]. Today, with the advents in technology, the prenatal diagnosis of this entity is possible. Here, we report the prenatal ultrasonographic features and diagnosis of a case with HI.

Case Report

A 23-year-old gravida 2 para 1 woman, at the 26th week of gestation, was referred to our perinatology unit for second opinion ultrasound (US). The couple was third degree consanguine and their first baby died on the fifth day of life, with the diagnosis of HI. Her personal and family history was otherwise unremarkable. In the present pregnancy, she did not report any medication use, had no history of fever (with or without rash), ionizing radiation exposure during the first trimester. First trimester screening for aneuploidy revealed a risk of 1:1818 for Down syndrome. Her routine pregnancy follow-up was eventless but she did not have a second trimester ultrasonographic examination for abnormality screening.

On 2-dimensional (2D), 3-dimensional (3D) and 4-dimensional (4D) US; polyhydramnios, intrauterine fetal growth retardation (EFW below the 10th percentile), facial dysmorphism with distorsion of the lips (eclabion), ectropion - conjunctival protrusion associated with severe chemosis, skin fissures, short digits, flat nose, severe edema on the dorsal surfaces of hands and feet were detected. These findings were considered as sonographic features of HI. (Figure 1-5). After proper counselling about the postnatal course of the disease and the risks of amniocentesis,

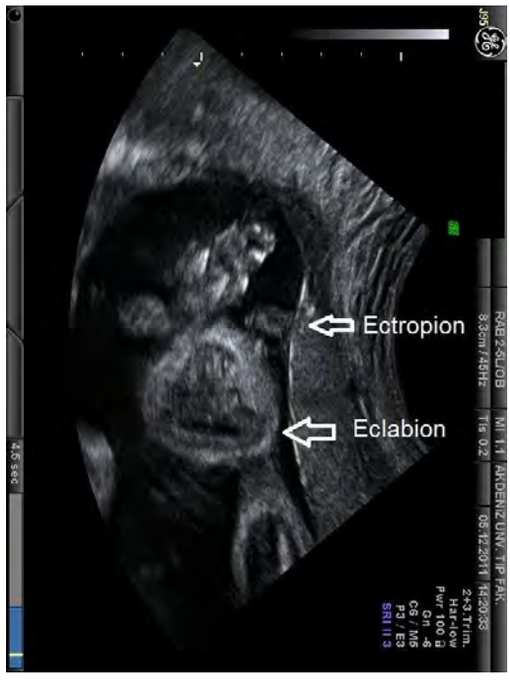

Figure 1. US view of ectropion and eclabion.

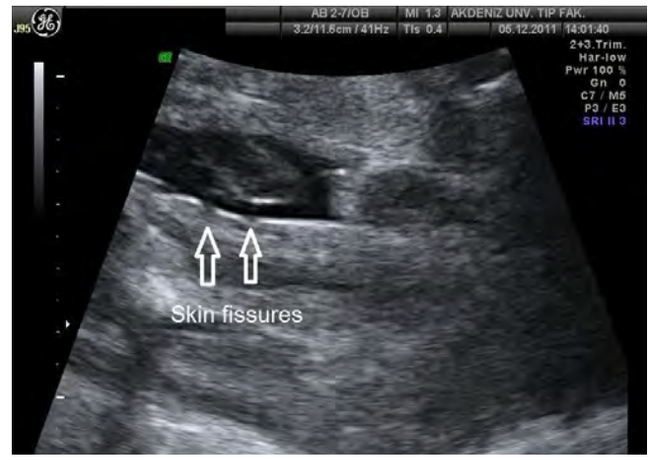

Figure 2. US view of skin fissures.

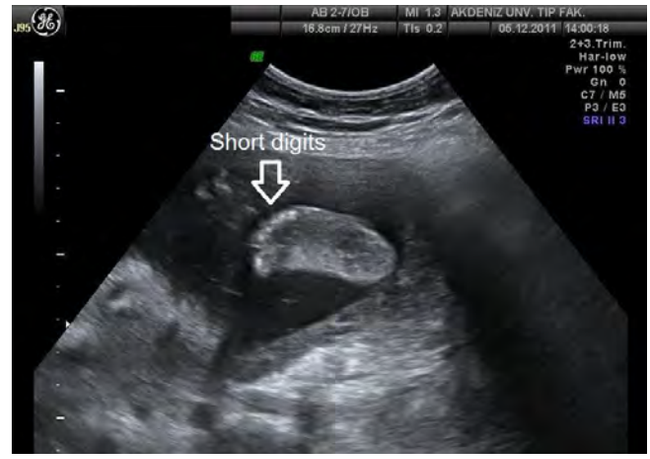

Figure 3. US view of short digits of foot.

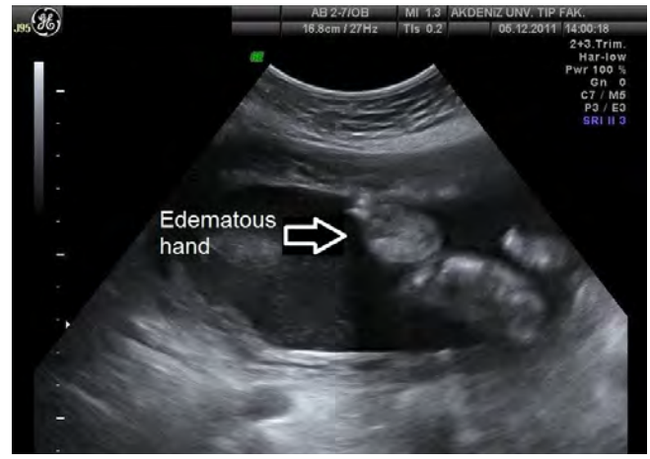

Figure 4. US view of edematous hand.

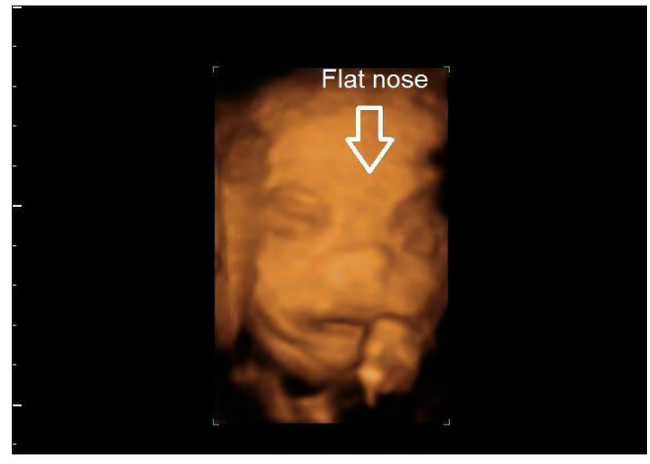

Figure 5. US view of flat nose.

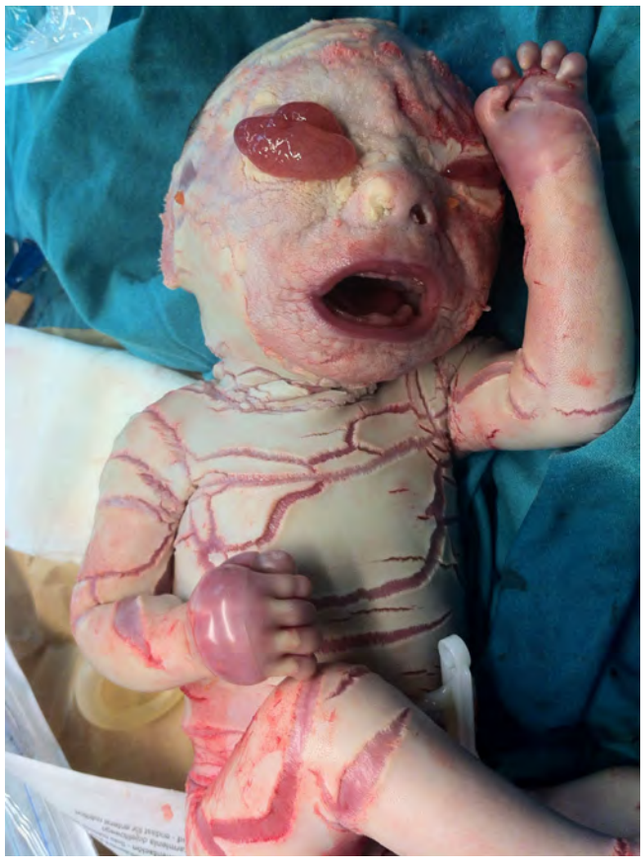

Figure 6. Postnatal features of HI. Note eclabion, ectropion, skin fissures and severe edema of hands.

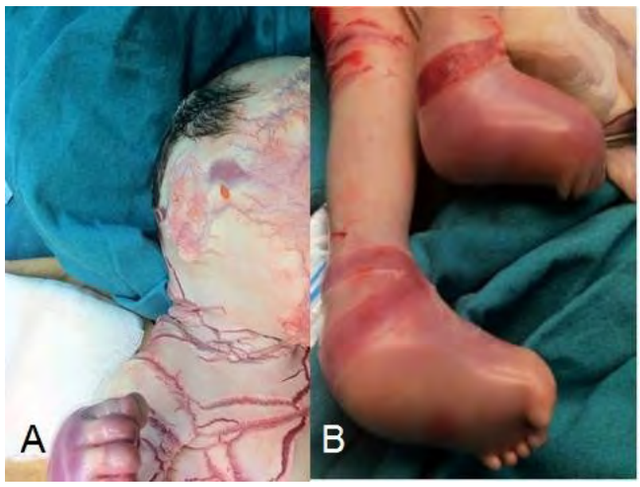

Figure 7. Poorly malformed ear (A). Edema of feet (B).

the parents denied to have any invasive procedures.

A male infant with HI (birth weight, 2130 gr) was delivered by cesarean section for breech presentation and preterm labor following premature rupture of the fetal membranes at 32 weeks of gestation. The infant manifested malformations, which were detected on antenatal US and additionally had poorly developed ears (Figure 6, Figure 7). He was hospitalized at the intensive care unit for 20 days and recieved regular application of emollients. The baby is now alive and 12 months old, receiving physiotherapy for extremities, routine pediatric and dermatologic follow-up and preventive care for infections and dehydration.

Discussion

HI is an overwhelming disorder and the phenotypical appearance of the neonate is devastating for both parents and health care providers. Therefore, early prenatal diagnosis of this disorder is particularly important.

Although it was previously reported that the inheritance of HI is autosomal recessive, in a large number of cases, the inheritance pattern cannot be ascertained, and the disorder could be due to a new dominant mutation [6]. Unfortunately, as in our case, autosomal recessively inherited disorders are relatively common probably secondary to high percentage of related marriages in Turkey.

To date, various ABCA12 mutations have been reported in HI patients [7]. Although some attemps were made to use DNA-based analysis for earlier (i.e., first trimester) prenatal testing [6], US and fetoscopic- or ultrasound-guided fetal skin biopsies are generally preferred for prenatal diagnosis. With the widely usage of 3D and 4D ultrasound, the characteristic suggestive features of HI as eclabion, ectropion, rudimentary ears, flexion contractures at the knees, and dense floating particles in the amniotic fluid, are reported to be detected with higher frequency [8-11]. Moreover, some promising therapeutic interventions based on targeted molecular therapy and gene therapy strategies are also defined [12].

In conclusion, prenatal diagnosis was relatively straightforward in our case due to recurrence, enabling targeted US. The US markers for HI should be kept in mind, particularly for early and accurate antenatal diagnosis of this condition.

Competing interests

The authors declare that they have no competing interests.

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Diplopia: A Rare Orthopedic Clinical Complaint After Knee Arthroscopy

Diz Artroskopisi Sonrası Ortopedi Polikliniğine Nadir Bir Başvuru Şikâyeti: Diplopi

After Knee Arthroscopy: Diplopia

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11 Türk Spor Yaralanmaları Artroskopi ve Diz Cerrahisi Kongresi 2012'de poster olarak sunulmuştur.

Özet

Otuz iki yaşında erkek hasta, on gün önce artroskopik diz cerrahisi için spinal anestezi aldıktan sonra başlayan, sol gözde diplopi (çift görme) şikâyeti ile polikliniğimize başvurdu. Hastanın konservatif takip sonrası ikinci haftada şikâyetlerinin azaldığı, birinci ay sonunda tamamen geçtiği gözlendi. Bu olgu sunumunda; spinal anestezi altında yapılan artroskopik diz cerrahisi sonrası, ortopedik cerrahların sık olarak karşılaşmadıkları diplopi şikayetiyle polikliniğimize başvuran olguyu tartışmayı amaçladık.

Anahtar Kelimeler

Diplopi; Abdusens Sinir Hasarı; Spinal Anestezi; Artroskopi; Komplikasyon

Abstract

The patient was a 32-year-old male who had undergone arthroscopic surgery under spinal anesthesia ten days prior to being admitted to hospital with a complaint of diplopia (double vision). The patient was examined and it was determined that the diplopia was due to sixth cranial nerve palsy. After conservative treatment, his complaints decreased after two weeks and completely resolved in one month. We present the case of diplopia due to spinal anesthesia after arthroscopic surgery, which is not a typical case seen by orthopaedic surgeons.

Keywords

Diplopia; Abducens Nerve Injury; Spinal Anesthesia; Arthroscopies; Complications

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Introduction

The most common complication after spinal anesthesia is headache and, more rarely, hearing loss, diplopia, tinnitus or complications, such as loss of consciousness, have been reported [1]. As a result of intracranial hypotension due to loss of cerebrospinal fluid (CSF), all intracranial nerves can be affected, except for intracranial nerves one, nine and ten. Due to the positioning of the nerves, the sixth intracranial nerve (N. abducens) is affected in 95% of the cases [2]. Diplopia occurs as a result of extraocular muscle paralysis with or without headache. It may be single-sided or double-sided.

In this case report, we have presented the case of diplopia seen 10 days after arthroscopic knee surgery in which the patient was administered spinal anesthesia.

Case Report

A 32-year-old male patient was admitted to hospital with complaints of diplopia (double vision). He had undergone arthroscopic left knee surgery under spinal anesthesia ten days prior to the admission for diplopia and he was discharged from the hospital the following day. Upon discharge, the patient was given information about how to prevent post-dural-puncture headache and he was warned to take fluids and get bed rest; however, despite these warnings, the patient stated that he did follow that advice or take the suggested measures. Two days after he was administered the spinal anesthesia, he experienced a headache that began in the back of his neck and changed position. Four days after undergoing spinal anesthesia, the patient began complaining about double vision. The patient had no additional disease and no history of drug use. He was consulted to the anesthesia department and hospitalised. The diagnosis was considered as sixth cranial nerve palsy due to the intracranial hypotension caused by cerebrospinal fluid loss during the spinal anesthesia. The patient's hematologic and biochemical values had been checked preoperatively and were interpreted as normal. Intraoperative and postoperative hemodynamic data from the anesthesia records were also interpreted as normal. The spinal anesthesia had been administered in a lateral position and the anesthesia had been placed into the intervertebral space to the left of the 3-4 lumbar vertebrae using a 25-gauge (G) pencil tipped needle by injecting 12.5 mg hyperbaric bupivacaine (Marcaine spinal injection of 0.5% heavy bulb, Astra Zeneca). The operation began 10 minutes after administration of the spinal anesthesia so that the patient's lower extremities were lateralised.

In the hospital, the patient's continuing headache severity was evaluated by a visual analogue score ranging from 1-2. Fluid replacement therapy was started with paracetamol + caffeine-containing analgesics and bed rest was recommended. Ophthalmology and neurology consultations were requested. During a right binocular diplopia examination, the ophthalmologist used the Maddox test and diagnosed diplopia due to the presence of right-sided sixth cranial nerve palsy. The patient's neurological examination was normal. Cranial MRI and diffusion MRI showed no pathological findings and visual field testing was normal. The patient noted that his diplopia had decreased during the treatment and, after four days, he was discharged upon his own request. The diplopia decreased during the second week and complete remission was seen one month later, during the patient's follow-up visit.

Discussion

Diplopia or extraocular muscle paralysis that occurs after dural initiatives often leads patients to visit the neurology and ophthalmology outpatient clinics. For diplopia, the rate of occurrence ranges between 1/400-1/8000 [3]. Diplopia occurs after spinal anesthesia in 47% of patients; it occurs after myelography in 18% of patients and it occurs after a lumbar puncture for diagnostic procedures in 18% of patients. The abducens nerve (sixth cranial nerve) is the cranial nerve that is most often affected and it is responsible for diplopia in 92% to 95% of all cases. Patients between the ages of 17-69 are typically affected, but this condition often occurs in patients who are older than 30 years of age. Abducens nerve palsy is more frequent in men according to Thorsen et al. [4], whereas other clinical reports note that it occurs equally among males and females [2]. Nerve abducens palsy is unilateral in 80% of cases [4]. Oculomotor (third cranial nerve) and trochlear (fourth cranial nerve) nerve involvement, although rare, can be added to the list of nerves that can be affected. It can be difficult to diagnose this condition in patients with more than one cranial nerve involvement [5].

Sixth cranial nerve palsy arises after or with a post-dural-puncture headache. Diplopia can occur after spinal anesthesia due to the loss of cerebrospinal fluid (CSF) leading to intracranial hypotension, which results in the displacement of cerebral structures. Due to the long intracranial course and the positioning of the nerve, the nerve becomes susceptible to mechanic damages. Prevention of CSF leakage is the most important way to reduce intracranial hypotension. Therefore, for spinal anesthesia, the use of thin gauge and blunt-tipped needles is recommended [6]. In our patient, we used a pencil tipped 25 G needle and it was a risk factor.

Diplopia can be observed one day to three weeks after spinal anesthesia. The onset of diplopia can often be seen between 4-10 days post-surgery in what is known as the 'window period' [4]. During this 'window period', patients often visit the neurologist or ophthalmologist instead of the orthopedic surgeon. Orthopedic surgeons are not aware of this complication because of its rare occurrence. The patient in this case study was the first patient at our clinic to complain about diplopia.

In the differential diagnosis of diplopia, malignancy, ischemia, trauma, aneurysm, multiple sclerosis, encephalitis and other causes must be ruled out [1]. Magnetic resonance imaging (MRI) is important in the differential diagnosis. Using magnetic resonance imaging, changes in pachymeningeal tissue, dural thickening and displacement of brain parenchyma due to the intracranial hypotension can be distinguished from other possible etiologic factors.

If there is isolated cranial nerve involvement and a history of prior headache and the diplopia begins three weeks after having undergone spinal anesthesia and no other neurological involvement is found, the developed diplopia can probably be interpreted as a complication of spinal anesthesia.

Post-dural-puncture headache treatment has both conservative and invasive treatment protocols. Conservative treatment

consists of bed rest, hydration, analgesics and the ingestion of caffeinated drinks [1]. The effect of bed rest after spinal anesthesia has not been proven to prevent the development of diplopia. Kose et al. [7] have reported the development of diplopia due to spinal anesthesia in a 38-year-old patient with hallux valgus. They decided that the use of conservative treatment is sufficient. Invasive therapy, such as an epidural blood patch, can be applied. The use of an epidural blood patch in the treatment of headache after spinal anesthesia has a 93% success rate, but this approach does not show the same success rate in the treatment of diplopia [8]. Post-dural-puncture headache or diplopia may occur in patients receiving an epidural blood patch. In such a case, an epidural blood patch may be responsible for the development of diplopia. On the other hand, in undiagnosed subdural hematoma after spinal anesthesia in patients with neurological symptoms, the use of an epidural blood patch can lead to increased incidences of symptoms [7].

Before using an epidural blood patch, a complete medical history should be taken. The patients should be clinically evaluated in terms of subdural hematoma. Due to the decrease in the severity of symptoms in our patient, conservative treatment was continued.

Abducens nerve palsy after spinal anesthesia often has a good prognosis and it often resolves completely at any point between four weeks and four months. If isolated abducens nerve palsy occurs in patients without evidence of other neurological symptoms, the progression of the condition should be monitored and diplopia should be treated conservatively.

The lateralisation in the left side position for 10 minutes can lead to the development of right-sided sixth cranial nerve palsy because the force of gravity leads to increased traction on the right side of the sixth cranial nerve. In the literature, we did not see the effect of lateralisation on the cranial nerve. However, the implementation of left lateralisation suggests that this mechanism may explain right-sided sixth cranial paralysis.

Conclusion

Outpatient arthroscopic knee surgery patients who were discharged should be advised to undergo bed rest and increase their fluid intake. In addition to headaches, diplopia can also be seen as a complication and information about preventing or reducing symptoms should be given to patients. When patients who have undergone postoperative orthopedic surgery with spinal anesthesia visit the clinic, complaining of diplopia, they should be asked about their medical history. It is important for the surgeon and the patients to know that diplopia caused by spinal anesthesia is reversible. This situation should not be a surprise to orthopedic surgeons.

Competing interests

The authors declare that they have no competing interests.

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Postpartum Pemphigoid Gestationis: Good Respons to Topical Clobetasol

Postpartum Gestasyonel Pemfigoid: Topikal Klobetazole İyi Cevap

Postpartum Pemphigoid Gestationis

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Özet

Gestasyonel pemfigoid(GP) nadir görülen gebelikle ilişkili otoimmun subepidermal büllöz bir hastalıktır. Genellikle ikinci ve üçüncü trimesterde görülmekle beraber nadiren birinci trimesterde ya da postpartum dönemde de ortaya çıkabilir. Polikliniğimize 36 haftalık normal spontan doğum sonrası onikinci günde karın, bacaklar ve kollarda kaşıntılı su toplayan yaralar şikayeti ile başvuran 25 yaşında kadın hastanın yapılan dermatolojik muayenesinde karın cildi, her iki uyluk mediali ve önkol fleksör yüzlerde eritemli zeminde gergin büller izlendi. Anne ve bebek diğer açılardan sağlıklı idi. Yapılan biyopsinin immunfloresan ve hematoksilen eozin incelemesi sonucu postpartum GP tanısı konulan hastaya topikal klobetazol propiyonat tedavisi başlandı. Hastanın lezyonları tamamen düzeldi. Hastamızı nadir görülen, gebelikle ilişkili otoimmun büllöz bir hastalık olan gestasyonel pemfigoidin postpartum dönemde ortaya çıkması ve topikal steroide iyi yanıt vermesi açısından, bildirmeyi uygun gördük.

Anahtar Kelimeler

Gestasyonel Pemfigoid; Postpartum Dönem; Klobetazol Propiyonat

Abstract

Pemphigoid gestationes is a rare pregnancy related autoimmune subepidermal bullous disease. It frequently occurs at second and third trimester of pregnancy. Also it rarely appears at first trimester of pregnancy or postpartum period. Twenty-five years old woman was admitted our outpatient clinic at 12 th day of delivery with distance bullous lesions over erythematous area at abdominal skin, medial site of thighs and flexor site of forearms bilaterally. She and her baby were healthy in other respect. She was diagnosed as pemphigoid gestationes after hematoxylin and eosin and direct immunofluorescence examination of biopsy and topical clobetasol propionate ointment was initiated topically twice daily. The lesions were regressed almost completely. We present this case because of its' infrequent situation of pregnancy related autoimmune bullous disease and good respond to topical steroid treatment.

Keywords

Pemphigoid Gestationis; Postpartum Period; Clobetasol Propionate

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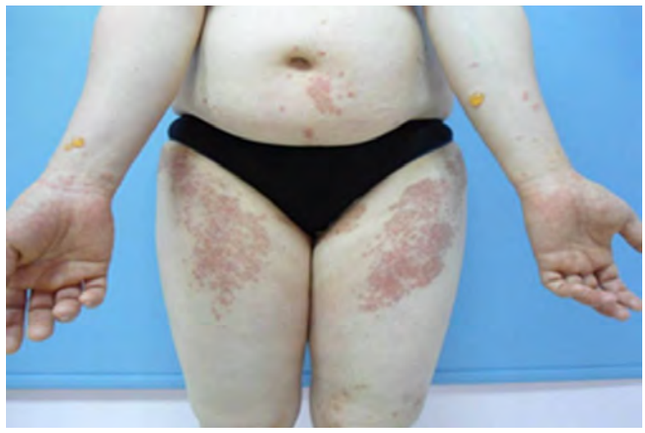
Giriş

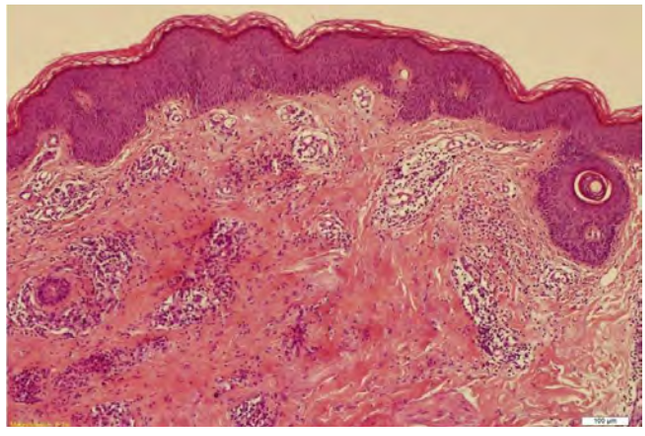
Gestasyonel Pemfigoid (GP) doğurganlık çağındaki kadınları etkileyen 50000'de bir görülen nadir immunobüllöz bir hastalıktır[1,2]. Çoğunlukla üçüncü trimesterde ortaya çıkmakla birlikte erken gebelik haftalarında da görülebilir. Tedavide ilk seçenek sistemik kortikosteroid tedavisidir. Bazı yazarlar erken postpartum dönemde kısa süreli steroid dozunu yükseltmeyi de önermektedirler. Bunun yanında dirençli olgularda siklosporin, azotioprin ve metotreksat önerilmektedir[3]. Hastalık doğum sonrası haftalar, aylar yada yıllar içinde iyileşme eğilimindedir[1]. Doğum sonrası başlayan GP olguları nadiren bildirilmiştir[4]. Burada postpartum ikinci gün gelişen ve topikal steroid tedavisine iyi cevap veren bir GP hastası sunulmaktadır.

Olgu Sunumu

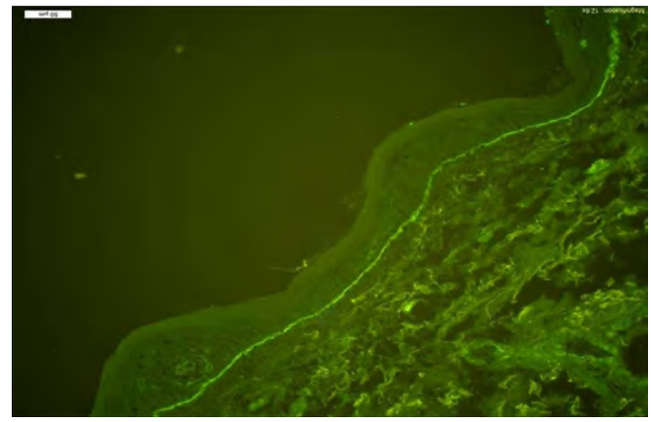
Yirmibeş yaşında bayan hasta vücudunda 10 gündür mevcut olan kaşıntı ve su toplaması şikayeti ile başvurdu. Hastanın öyküsünde 12 gün önce 36. gebelik haftasında normal spontan doğum yaptığı öğrenildi. Dermatolojik muayenesinde karın, kol ve bacakların fleksör yüzlerini tutan, eritemli plaklar üzerinde gergin büller izlendi (Resim 1). Avuç içi ayak tabanı ve mukozalar korunmuştu. Hastanın genel durumu iyi ve sistemik muayenesi normaldi. GP, büllöz pemfgoid, lineer Ig A dermatozu ve eritema multiforme ön tanıları ile alınan biyopsisinin histopatolojik incelemesinde üst ve orta dermiste peri vasküler lenfoplazmositik hücre infiltrasyonu ve eozinofiller izlendi (Resim 2). Direk immunfloresan incelemede bazal tabakada belirgin C3 ve hafif Ig G birikimi görüldü (Resim 3). Bu bulgular ışığında postpartum GP tanısı konularak lezyonların şiddetli olmaması nedeniyle hastaya günde iki kez topikal %0,05 klobetazol propiyonat merhem ve günde bir kez sistemik antihistaminik olarak setirizin hidroklorür 10 mg tedavisi başlandı. Tedaviyle lezyonları gerileyen hastanın ilk 3 ay az sayıda lezyon çıkışı olmakla beraber günde iki kez uygulanan topikal klobetazol propyonat ile bu lezyonlar da geriledi. Takipte postpartum altıncı ayda yeni lezyon çıkışı yoktu.



Resim 1. Karın, önkol ve bacakların fleksör yüzlerini tutan eritemli plaklar üzerinde yer yer gergin büller.



Resim 2. Üst ve orta dermiste peri vasküler lenfoplazmositik hücre infiltrasyonu ve eozinofiller (Hematoksilin eozin X 20)



Resim 3. Direk immunfloresan incelemede bazal tabakada belirgin C3 ve hafif Ig G birikimi

Tartışma

Gestasyonel pemfigoid nadir görülen gebelikle ilişkili otoimmun subepidermal büllöz bir hastalıktır [1]. Genellikle ikinci ve üçüncü trimesterde görülmekle beraber nadiren birinci trimesterde ya da postpartum dönemde de ortaya çıkabilir [5,6]. Nadiren koryokarsinoma ve mol gebelikle birlikte görülebilir [4]. Plasental MHC II antijenlerinin anormal ekspresyonu immünolojik olayların başlamasından sorumlu tutulmaktadır. Bazal membran yapılarına karşı antikor oluşumu söz konusudur. PG'te hedef antijen, 230 kd BPAg1 ve 180 kd BPAG2'dir. GP patogenezinde plasentada ve deride bazal membranda bulunan kollajen XVII'ye (BP Ag 2 (180 kd BP Ag)) karşı gelişmiş antikorlar rol oynar. İmmunogenetik çalışmalar GP ile HLA DR2, HLA DR4 doku uygunluk antijenlerinin birlikteliğini göstermiştir[1,6].

Postpartum GP olguları çoğunlukla gebelikte başlayan hastalığın alevlenmesi şeklindedir. Doğum sonrası başlayan GP olguları nadiren bildirilmiştir [5]. Postpartum GP sıklıkla doğum sonrası saatler içinde başlar. Doğumdan 3 ve daha fazla gün sonrasında başlaması ise nadirdir. Sonraki gebeliklerde, menstruel siklus esnasında ya da oral kontraseptif kullanımında nüksler görülebilir[6]. Bizim olgumuzda ise lezyonlar doğumdan 2 gün sonra başlamıştı.

GP'de lezyonlar göbek çevresinden başlayarak yayılır. Nadiren avuç içi ayak tabanı, mukoza tutulumu görülür. Olgumuzda palmoplantar tutulum ve mukozal tutulum görülmedi.

GP için belirlenmiş tanı kriterleri klinik, histopatoloji, immunfloresan ve laboratuvar bulgulardan oluşur[7] (Tablo 1). Histopatolojik bulgular klinik bulgularla uyumlu olarak çok geniş bir yelpazede görülebilir. Ürtikeryal lezyonlar söz konusu olduğunda eozinofilik spongiyoz, dermal lenfohistiyosit ve eozinofil infiltrasyon, papiller dermal ödem, vezikül ve bül söz konusu olduğunda ise dermoepidermal ayrışma, bazal membranda nekroz ve dermal mikst inflamatuar infiltrat görülür. Castro ve arkadaşlarının 10 hastalık serisinde sadece 3 hastada histopatolojik olarak

Tablo 1. Gestasyonel pemfigoid için tanı kriterleri

Klinik	Gebelik sırasında ya da hemen doğum sonrası ortaya çıkması Kaşıntılı ürtikeryal papül, plaklar beraberinde büller görülmesi Göbek çevresinden başlaması
Histopatoloji	Eozinofilik spongiyoz Subepidermal büller
DIF	Eozinofil ve lenfositlerden oluşan dermal infiltrat
IIF	Dermoepidermal alanda lineer IgG C3 birikimi Tuzla ayrıştırma tesitinde birikim epidermal alanda Bazal membrana karşı dolaşımda IgG antikorları
ELİSA	Kollojen XVII'ye karşı dolaşımda IgG antikorları
İmmunbloting	Dolaşımda 180 kd yada 230 kd protein bandında IgG antikorları varlığı
HLA	HLA DR 3, DR4

dermoepidermal bül oluşumu gösterilmiştir[8]. Bizim olgumuzda biyopsi ürtikeryal alandan alındığı için dermoepidermal ayrışma izlenmedi, buna karşılık dermal perivasküler lenfohistiosit ik ve eozinofilik infiltrasyon izlendi. GP 'de immunfloresan incelemede C3 birikimi hastaların %100'de görülürken, Ig G birikimi %25-50 oranında görülür[3]. Hastamızın biyopsisinin immunfloresan incelemesinde iki pozitif C3 birikimi ve hafif Ig G birikimi izlendi.

GP tedavisinde birinci basamak 0,5 mg/kg dozunda sistemik steroiddir. Erken ürtikeryal lezyonlar topikal steroid ve sistemik antihistaminik tedavisine iyi cevap verir. Alternatif tedaviler dapson, sülfapridin, siklosporin ve pridoksini içerir. Dirençli olgularda adjuvan tedavi olarak metotreksat, azotioprin, altın, siklosporin ve pridoksin kullanılabilir[3,7]. Patsatsi ve arkadaşları tarafından, sistemik steroid tedavisine dirençli bir postpartum GP olgusunda plazmafrez tedavisine yanıt alındığı bildirilmiştir[9]. Cozzani ve arkadaşlarının bildirdiği postpartum GP olgusunda ise topikal steroid ve sistemik antihistaminik tedavisine iyi cevap vermiş[10]. Bizim olgumuz da diğer olgu gibi postpartum başlamasına ve ürtikeryal lezyonlara büllerin eşlik etmesine rağmen topikal steroid ve sistemik antihistaminik tedavisine iyi cevap verdi. İlk üç ay tek tük yeni lezyon çıkmakla beraber klobetazol propiyonat topikal uygulaması ile bunlar da geriledi. Postpartum 6. ayda hastamızda nüks olmadı.

Sonuç olarak gebelik dermatozları içinde yer alan PG postpartum dönem de de karşımıza çıkabilen otoimmun büllöz bir hastalıktır. Tedavide sistemik steroid birincil tedavi olmakla berber topikal yüksek potent steroide cevap veren olgularda vardır. Hafif ve orta şiddetli olgularda yüksek potensli topikal steroidler de tercih edilebilir. Hastalar oral kontraseptif kullanımı, menstruasyon ve tekrarlayan gebeliklerinde nüks açısından takip edilmelidir.

Çıkar Çakışması ve Finansman Beyanı

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Autosomal Dominant Polycystic Kidney Disease Patient Specified Bilateral Renal Mass: A Case Report

Otozomal Dominant Polikistik Böbrek Hastasında Belirlenen Bilateral Renal Kitle: Olgu sunumu

Autosomal Dominant Polycystic Kidney Mass of Bilateral

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Özet

Böbreğin kistik lezyonları herediter ve nonherediter kistleri içeren çok geniş bir lezyon aralığına sahiptir. Bunlar böbrek kisti şeklinde tek bulgu olabileceği gibi ekstrarenal bir klinik sendromun parçası da olabilir. Bu nedenle kistik lezyonlar değişik birimleri içeren multidisipliner yaklaşımı gündeme getirmektedir. Otozomal dominant polikistik böbrek hastalığı, her iki böbrekte farklı boyut ve sayıda kistik değişiklikler şeklinde kendini gösteren sistemik ve kalıtsal bir hastalıktır. Herediter ve bazı edinsel kistik hastalıklar zemininde tümörle birlikteliği çeşitli çalışmalarda ortaya konsa da nadirdir. Burada otozomal dominant polikistik böbrek hastalığı tanısı almış, son altı aydır batında ağrısız kitle nedeniyle yapılan fizik muayene, radyolojik ve laboratuvar tetkikleri sonucu bilateral renal kitle tanısı konan ve nadir görülen, sağ renal kitleye nefron koruyucu cerrahi, sol renal kitleye aktif izlem ile takip edilen hasta güncel literatür taranarak tartışılmıştır.

Anahtar Kelimeler

Polikistik Böbrek; Renal Hücreli Karsinom; Nefron Koruyucu Cerrahi

Abstract

Hereditary cystic lesions of the kidney cysts nonherediter lesion with a wide range. They may be the only finding in the form of extrarenal renal cysts may also be part of a clinical syndrome. For this reason, multi-disciplinary approach brings with cystic lesions in the different units. Autosomal dominant polycystic kidney disease, kidney different sizes and numbers of both systemic and hereditary disease that manifests itself in the form of cystic changes. Hereditary and acquired cystic lesions on the basis of some of the rare tumor association is determined in several studies. Here are diagnosed with autosomal dominant polycystic kidney disease, abdominal pain because of a mass in the last six months, physical examination, radiological and laboratory findings were diagnosed with bilateral renal tumors are uncommon and the right renal mass nephron-sparing surgery, the patients with left renal mass followed with active surveillance discussed in the current literature.

Keywords

Polycystic Kidney; Renal Cell Carcinoma; Nephron-Sparing Surgery

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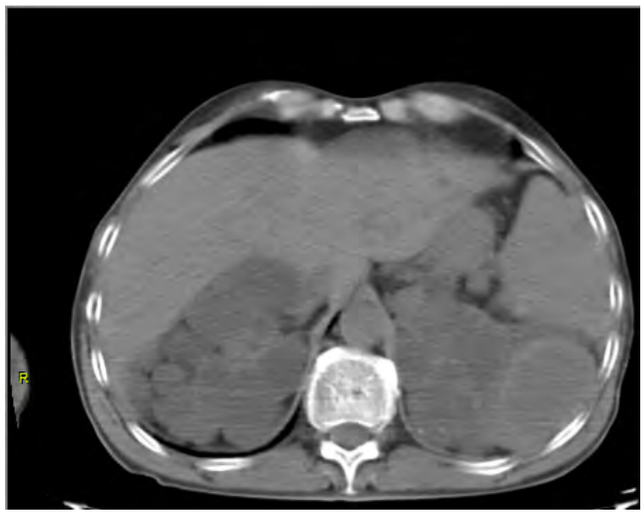
Giriş

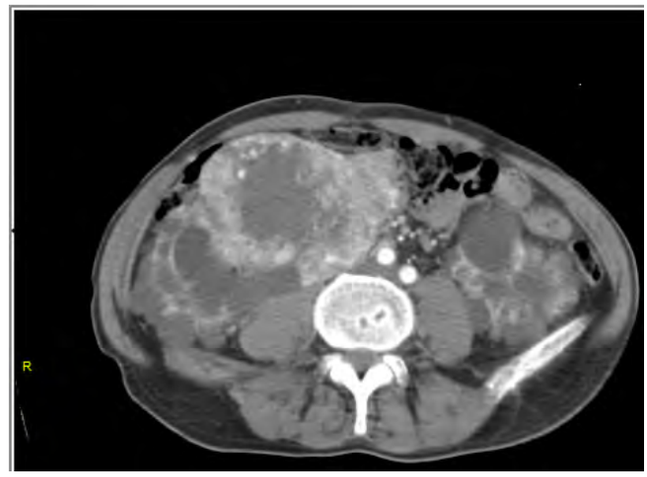
Kistik böbrek lezyonları; kalıtsal ve edinilmiş böbrek kistlerini ve kistik böbrek tümörlerini içerir. Otozomal dominant polikistik böbrek hastalığı en yaygın görülen kistik hastalık olup 4.000-10.000 doğumla birliktedir. Kistik böbrek hastalığı genellikle herediter ve gelişimsel nedenlere bağlı, böbrekleri diffüz ve bilateral olarak tutan, farklı boyut ve sayıda kistik değişikliklerle karakterizedir [1]. Böbreğin kistik hastalıklarından bazılarında böbrek tümörü oluşmadığı bilinse de otozomal dominant polikistik böbrek hastalığı ve böbreğin edinilmiş kistik lezyonları ile birlikteliği olabilir ancak nadirdir. Kistler karında kitlesel görüntüye ve metabolik olarak halsizlik ve yorgunluk yakınmasına neden olsa da spesifik olmayıp tümöral kitleler de bu yakınmaya neden olabilirler [2].

Olgu Sunumu

Elli sekiz yaşında otozomal dominant polikistik böbrek hastalığı tanısı konan ve takip altında olan hasta son altı aydır ağrısız, karında bombeliğe neden olan şişlik, kilo kaybı ve iştahsızlık nedeniyle başvurdu. Hastanın 6 yıldır herediter polikistik böbrek tanısı var olduğu ve aile taramasında diğer bireylerde de tespit edildiği öğrenildi. Fizik muayenesinde sağ alt kadrana uzanan, ön duvarda bombelik oluşturan, sert nodüler kitle belirlendi. Hastanın idrar tetkikinde önemli bir sorun yoktu ve idrar kültüründe üreme olmadı. Kan tahlilinde hafif anemisi vardı. Böbrek fonksiyon testlerinde bozulma tespit edilmedi ve dialize girmediği öğrenildi. Karaciğer fonksiyon testleri ve kanama pıhtılaşma zamanı normaldi. Tüm batın ultrasonda her iki böbrekte büyüğü 14x9 cm boyutlarında multipl kistik lezyon; sağ böbrek alt kesimde 14x9 cm, sol böbrek üst kesimde 8x5.5 cm ve sol böbrek alt kesimde 5x3.5 cm boyutlu solid kitle lezyonları tespit edildi. Akciğer grafisi normaldi. Tüm abdomen BT' de her iki böbrekte ve karaciğerde çok sayıda ve farklı boyutlarda kistik lezyonlar vardı (Resim-1). Sağ böbrekte 11.5x8.5 cm boyutunda kontrast tutan, santrali nekrotik özellikte solid kitle lezyonu, sol böbrek üst ve alt polde 4.5 cm çaplı kontrastlanma gösteren komplike kistik lezyon ve sol böbrek orta kesimde 1 cm büyüklüğünde kontrast tutan solid kitle lezyonu izlendi (Resim-2). Sağ böbrekteki kitle için nefron koruyucu cerrahi; sol böbrekteki komplike kist ve solid kitleye yönelik perkütan radyofrekans ablasyon ve/veya krioterapi, bir alternatif olarakta aktif izlem tedavi seçenekleri önerildi. Hastaya ameliyat ile ilgili bilgi verildi ve onam formu dolduruldu. Genel anestezi altında supin pozisyonunda orta hat; ksifoidden suprapubik bölgeye kadar uzanan kesi ile kitleye ulaşıldı ve sağ nefron koruyucu cerrahi ile kitle çıkartıldı. Ortama bir adet dren konarak anatomiye uygun olarak insizyon hattı kapatıldı. Hastanın postop birinci gün sorunu yoktu ve drenden akıntı gelmemesi üzerine alındı. Biyokimyasal testleri normal olan hasta dialize ihtiyaç duymadı ve postop 3. gün şifa ile taburcu edildi. Kitle histopatolojik olarak incelendi ve renal hücreli karsinom olarak raporlandı. Postoperatif birinci, üçüncü ve altıncı aylarda kan sayımı, biyokimyasal testler ve ultrasonografi yapıldı ve sol böbrekteki kitlelere perkütan yaklaşımları kabul etmemesi üzerine Bosniak Tip 4 kistler ve sol böbrekteki 1cm çapındaki solid kitle için aktif izlem kararı alındı. Bir yıllık kontrol vizitleri esnasında solid kitlede boyut artışı saptanmadı. Böbrek fonksiyon testleri normaldi ve diyalize ihtiyaç duyulmadı.



Resim 1. Her iki böbrekte çok sayıda ve boyutta kist



Resim 2. Sağda 11.5x8.5 cm ve sol böbrekte 1 cm kontrast tutan kitle

Tartışma

Otozomal dominant polikistik böbrek hastalığı en yaygın kalıtsal hastalıklardan biri olup yaşamın herhangi bir evresinde görülse de en sık 2. ve 3. Dekadda ortaya çıkmaktadır. Böbrek ile birlikte beyin, karaciğer, kardiyo-vasküler, gastro-intestinal ve kas-iskelet sistem tutulumu da görülebilir [3]. Başlangıçta boyut olarak küçük kistik yapıda iken, büyük boyutlara da ulaşan lezyonların tanı ve takibinde ultrasonografi kullanılabilir [4]. Kistin duvarında gelişebilecek kalsifikasyonları belirlemek, beraberinde gözlemlenebilen böbrek ve üreter taşlarını tespit etmek için kontrastsız BT tercih edilmelidir [5,6]. Yüksek doku çözünürlüğü nedeni ile kanamalı kistleri belirlemede ve allerji öyküsü olan hastalarda MR görüntüleme BT' ye tercih edilebilir. İnce kesitlerle yapılan renal tomografi hala böbrek tümörlerinin karakterini belirlemede kullanabileceğimiz en önemli radyolojik tetkiktir. Genel olarak BT' de ya da MRG' de kontrast tutan bütün kitleler aksi ispat edilene kadar renal hücreli karsinom olarak değerlendirilmelidir [7]. Otozomal dominant polikistik böbrek hastalığında böbrek-hücre proliferasyon risk oranı artmıştır. Hücre proliferasyonu sonucu epitelyal hiperplazi, adenom ve böbrek adenokarsinomu gelişebilir. Otozomal dominant polikistik böbrek hastalığı tanısı konan genç yaştaki hastalarda tümör bulguları ile birlikte, multifokal ve sarkomatoid yapı içermesi daha agresif seyirli tümör gelişmesi sıktır. Histopatolojik olarak en

sık berrak ve papiller hücreli tip gözlenirken, sarkomatoid komponent içerebileceği akılda tutulmalıdır [2]. Herediter kist tanısı almış hastalar genelde kist boyutunun artması ile birlikte özellikle kaşektik yapılı hastalarda daha belirgin olmak üzere karın ön duvarına bası yapan kitle bulgusu verebilir ancak spesifik dğildir. Bu hastalar tekrarlayan üriner taş ve kist boyutunun artması ile birlikte basıya bağlı lomber ağrı ve üriner enfeksiyon tablosu ile gelebilirler. Polikistik böbrek hastalığının tanı aşamasında ve takibinde ultrason bize değerli bilgiler verse de kitle tespiti halinde kitlenin özelliğini belirleme aşamasında yetersiz kalabilir. Bu aşamada tercih edilmesi gereken başlıca yöntem kontrastlı BT' dir. Olgumuzda ultrasonografi ile tespit edilen komplike kistin ve solid lezyonların BT' de kontrast tutuyor olması, lezyonların natürü ve evrelendirilmesi açısından önemini tartışmasız hale getirmektedir. BT ile ortaya konan ve kontrast tutan kitleler aksi ispat edilene kadar renal hücreli karsinom olarak değerlendirilmelidir. Sonuç olarak otozomal dominant polikistik böbrek hastalarında böbreklerde kist dışı kitlesel lezyonlar nadirde olsa gelişebilmektedir. Kitleyi değerlendirmek için ultrason önemli bilgiler verebilir ancak kontrastlı tomografi vazgeçilmezdir. Kontrast tutulumu olan kitleler aksi ispat edilene kadar malign tümör kabul edilmeli ve seçilmiş vakalarda nefron koruyucu cerrahi ve minimal invaziv tedavi yaklaşımları ile aktif izlem seçeneği hastaya sunulmalıdır.

Çıkar Çakışması ve Finansman Beyanı

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Abruptio Placenta Caused by Gunshot Injury in Second Trimester: A Case Report

İkinci Trimesterde Ateşli Silah Yaralanmasına Bağlı Oluşan Plasenta Dekolmanı: Bir Olgu Sunumu

Gunshot and Abruptio Placenta

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Özet

Yirmi dokuz yaşında multigravid bir gebe ateşli silah yaralanması nedeniyle kliniğimize başvurdu. Anamnezde 24 haftalık gebeliği mevcuttu. Yapılan ultrason muayenesinde retroplasental yaklaşık 6 cm lik hipoekoik bir alan ve bu bölgeye yakın yaklaşık 1 cm lik hiperekojen odak izlendi. Yine batın inspeksiyonunda umbilikusun 2 cm altında kanayan bir kesi alanı izlendi. Dekolman plasenta tanısıyla acil cerrahi düşündük. Ameliyat esnasında plasentanın % 50 dekole olduğu ve plasenta ile myometrium arasında bir mermi parçası olduğunu gördük. Mermi parçasını çıkartıp uterusun perfore alanı ve sezaryen kesi alanını tamir ettik. Ateşli silah yaralanmasının anne ve fetusu etkilemeksizin sadece abrusyo plasentaya yol açması nedeniyle bu vakanın önemli olduğunu düşündük.

Anahtar Kelimeler

Ateşli Silah; Gebe; Uterus

Abstract

A twenty-nine year old, multigravida woman was admitted to our clinic due to a gunshot injury. She had 24 weeks of gestation. It was observed by ultrasonography that there were 6 cm hypoechogenic image retroplacentally located and 1 cm hyperechogenic focus near to this image, and there was also a 2 cm actively bleeding incision area in subumbilical region by inspection of abdomen. We decided emergent operation because of the diagnosis of abruptio placenta. During the operation, we observed a piece of bullet between the placenta and myometrium and 50% separation of the placenta. We removed the piece of bullet, repaired the uterine perforation and then performed a caesarean section. We considered that this case is important because a gunshot injury had only caused an abruptio placenta without affecting mother and her fetus seriously.

Keywords

Gunshot; Pregnant Woman; Uterus

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Introduction

Abruptio placenta (AP) is defined as the preterm partial or complete separation of normally implanted placenta from the uterine wall [1]. The incidence of 4.4% has been reported in developing countries. Although the etiology is generally multifactorial maternal trauma may be an important reason [2]. Trauma is the leading nonobstetric cause of maternal death. Penetrating trauma during pregnancy primarily involves gunshot and stab wounds. The incidence of visceral injury in pregnant women with penetrating abdominal trauma is 16% to 38% versus 80% to 90% in the general population [3]. Gunshot wounds to the abdomen during pregnancy are becoming increasingly common, especially in countries adjacent to those impacted by war.

There is extensive literature about gunshot wound in pregnancy influencing the fetus, rectosigmoid junction, and the jejunum [4,5]. In our case, only AP was observed and had not affected any part of the fetus, membranes, bowel, and other maternal abdominal organs. Therefore, this case was considered rare, interesting and important.

Case Report

A twenty-nine year-old multigravida woman whose gravity 4, parity 3, was admitted to our clinic due to a gunshot injury. She had 24 weeks' gestation. In an ultrasound examination, it was found that the fetus was alive but bradycardic, there was a 6-cm hypoechogenic area located retroplacentally and a 1-cm hyperechogenic focus near to this area. Also in fetal examination; femur length was 22 weeks, biparietal diameter was 22 weeks- 1 day, fetal weight was 390 g. At the inspection, subumbilical a 2-cm actively bleeding incision was revealed. On vaginal examination, the cervix was closed and there was no vaginal bleeding.

On admission to our hospital the patient was hemodynamically stable. The initial Hb was 10.7 g/dL; Hct 34.8%; WBC 12.700; Plt 244.000; APTT 24.1; INR 0.96; blood pressures 110/85 mmHg; and heart rate 90 beats per minute.

We decided to make an emergent surgery due to AP after the family's informed consent. An emergent caesarean section under general anesthesia was performed. During the operation, we observed a 2-cm bleeding rupture area in the uterine fundus, peritoneal cavity contained approximately 300 ml of coagulated blood and 50% separation of the placenta after fetal delivery (figure 1). The fetus was born 350 g, a single male and intact but APGAR score was 0-0. We found a piece of bullet between the placenta and myometrium (figure 2). We removed a piece of bullet, then repaired the uterine perforation and performed a caesarean section. A general surgeon was consulted intraoperatively to evaluate her intestine, liver, spleen and other organs. There was no additional pathological finding. The neonatologist resuscitated but the fetus didn't respond to resuscitation. The metallic object was given to the hospital police, noted in the official report (figure 3).

There were no complications postoperatively. The woman was discharged four days after the caesarian section.

Discussion

Abruptio placenta (AP) is defined as the preterm partial or complete separation of a normally implanted placenta from

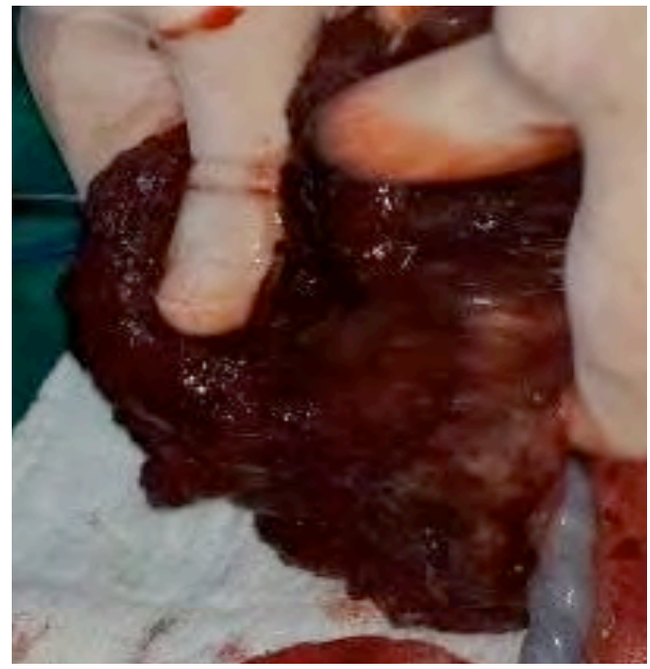

Figure 1. Placental abruptio area

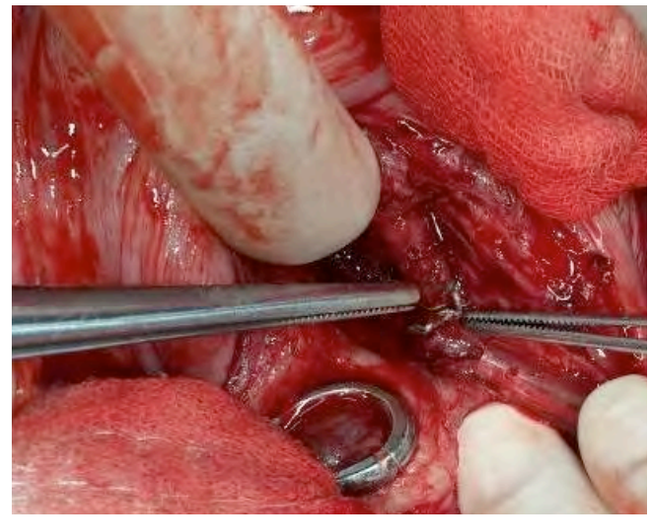

Figure 2. Metallic piece of bullet embedded to myometrium, after placental removal

Figure 3. Metallic piece after the operation

the uterine wall [1]. Etiology is generally multifactorial, some of these are placental insufficiency, intrauterine hypoxia, hypertensive disorders of pregnancy, nonvertex presentation, polyhydramnios, advanced maternal age, cigarette smoking, short umbilical cord, sudden decompression of the uterus, retroplacental fibromyoma, retroplacental bleeding from needle puncture (i.e. post amniocentesis), prior fetal demise, previous miscarriage, grand multiparity, preterm rupture of membranes, low prepregnancy body mass index and maternal trauma [1,2].

Motor vehicle accidents are the most common cause of nonobstetric trauma, and account for up to 80% of trauma in pregnancy [6]. Penetrating trauma has been found to be the cause of 9% of all pregnant trauma admissions. Of those, 73% was gunshot wounds, 23% stab wounds and 4% shotgun-related. Penetrating trauma in pregnancy was associated with a maternal mortality of 3.9-7% and with an increased fetal mortality (as high as 73%) [7].

Gunshot wounds to the abdomen during pregnancy are becoming increasingly common. In this case AP had been caused by trauma due to a gunshot. The literature about trauma during pregnancy indicates that 4-8% of women with trauma are sufferred from their close partners [8,9]. In our case, the woman did not know who guilty was because she was exposed to this accident while attending the wedding of her neighbor. It may be battle bullet from Syria where is close to the wedding place or by the any one of the people in the wedding.

An important fact is that pregnant women, who is sufferred from trauma, are special patients because pregnancy causes physiological and anatomical changes: such as increased blood volume, an expansion of plasma volume, which causes a dilutional effect known as physiological anemia and increased heart rate. These changes allow a loss of up to 30 % of circulating volume without changes in vital signs; however, the fetus may be affected. Management of these patients should be multidisciplinary, by the general surgeon, the obstetrician and the neonatologist [5]. In our case the patient was hemodynamically stable but the fetus was born 350 g, a single male and APGAR score was 0-0, so the neonatologist resuscitated but the fetus didn't respond to resuscitation.

Advanced imaging techniques (i.e. MR, CT) in traumatic patient can be done if the patient is stable condition, however ultrasound is often easily accessible in an emergency department and can provide crucial information in the pregnant patient with trauma [10]. AP is principally an emergency condition and might proceed agressively hypovolemic shock, disseminated intravascular coagulation and maternal death if the patient is not treated immediately. So we performed only ultrasound, although our patient was hemodynamically stable when she was admitted.

The gravid uterus is a protective barrier against other organs during trauma, so it is more likely to be injured. A great amount of energy of the trauma is absorbed by the uterine musculature, amniotic fluid and the fetus. This reduces the force of the impact and the possibility of damaging other organs. The results in the fetus are generally worrisome, causing direct impact up to 60-90%. In these cases maternal mortality is around 7-9% while fetal mortality is approximately 70% [7,11]. In our case, the uterus was in supraumbilical level and had absorbed the piece of bullet, therefore, other intraabdominal organs had remained intact. Although the fetus is intact the gestational week was low, 24 week, and AP was occurred. Because of this, pregnancy loss was inevitable.

Conclusion

Gunshot wounds are becoming increasingly common in society. The pregnant women is also at great risk. Often the mother and fetus are both affected. Less seldom, they both are unaffected but the placenta may be affected. So, an ultrasound examination should be done carefully in all traumatic pregnant women. In the management of a pregnant woman with a gunshot injury, it must be considered from three perspectives; the mother, the fetus and the placenta.

Competing interests

The authors declare that they have no competing interests.

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Fixator-Assisted Lengthening and Deformity Correction Over an Intramedullary Nail in a Patient with Achondroplasia

Akondroplazili Hastada Eksternal Fiksatör ile Çivi Üzerinden Uzatma ve Deformite Düzeltme

Lengthening and Deformity Correction in a Patient with Achondroplasia

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*This study is presented as a poster presentation at the National Orthopedics and Traumatology Congress in 2015.

Özet

Akondroplazi orantısız boy kısalığının en sık görüldüğü rizomelik kısalık formudur. Hastalar günlük yaşamda büyük oranda fiziksel ve psikolojik sorunlarla karşılaşırlar. Akondroplazili hastalarda boy uzatma ve deformite düzeltme birçok farklı metodla yapılabilir. Bu vaka sunumunda, 17 yaşındaki akondroplazili bayan hastada iki taraflı alt ekstremite uzatma ve iki taraflı tibial varus deformitesi düzeltme tedavisi sunulmaktadır.

Anahtar Kelimeler

Akondroplazi; Deformite; Kısalık; Düzeltme; Uzatma

Abstract

Achondroplasia is the most frequently encountered form of nonlethal skeletal dysplasia and a type of rhizomelic dwarfism. It results in considerable physical and psychologic handicaps owing to the disproportionate stature of the body and difficulty in performing routine activities of daily living. They also have major musculoskeletal problems including symptomatic malalignment of the lower limbs. Limb lengthening has been used in patients with achondroplasia by different techniques (Intramedullar nailing, monolateral or circular external fixator). We report our treatment of a patient 17 years of age with achondroplasia for bilateral lower limb length discrepancy and bilateral tibial varus deformity.

Keywords

Achondroplasia, deformity, shortness, correction, leghthening

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Introduction

Achondroplastic dwarfism results in considerable physical and psychologic handicaps and difficulty in performing routine activities of daily living [1]. These individuals often suffer from emotional disturbances, and are prone to have inferiority complexes [1]. They also have symptomatic malalignment of the lower limbs. Some patients eventually undergo serious surgery for correction of their malalignment to decrease pain and prevent early-onset degenerative arthritis [2,3]. Lower limbs can be lengthened with chondrodiastasis (the growth plate distraction) and callotasis (the callus distraction) successfully [4]. Nowadays in common practice both the tibia and femur are simultaneously lengthened in different sessions [6]. In this study, We report the result of using a combination of fixator-assisted nailing with lengthening over an intramedullary nail in a 17 year old patient with achondroplasia with tibial deformity and shortening.

Case Report

17 years old patient referred for leg lengthening for cosmetic purposes and difficulty of walking due to bilateral tibial varus deformity. Anterior posterior and lateral radiography of both legs were taken to determine the calf length, extremity length, the appropriate nail characteristics and the tibial varus angle. The varus deformity was 18° for both tibias and extremty length for both sides was 59 cm. At the initial radiologic examination there were Harris Muller plates in both proksimal femurs from an operation that she had at two years of age (Figure 1). We notified the patient and her family about the complete procedure, the possible complications as well as the duration of the process. Informed consent was obtained from the patient and her family. We planed to operate extremities as a cross in different sessions. Firstly we removed both plates and lengthen the right femur with intramedullar nailing with fixator assisted method; and the contralateral tibia with the same method correcting the tibial varus deformity (Figure 2). For the right femur we used intrammedullar nail (IMN) and because of the small calf of the tibia two of 3 mm IM elastic nails were used for tibial fixation (Figure 2). Under general anestesia, on supin position; from the proximal enterence the right femur was reamed of the intramedullary canal over an intramedullary nail (of the diameter of 10 mm, 260 mm length). Then osteotomy for femur was made from the subtrochanteric region and maximum possible size of nail was performed regarding the length of femur. After locking the femoral nail distally, monolateral external fixator was used; two pins were inserted proximally and three for distally. For the tibial deformity of contralateral side, after inserting two intramedullary elastic nails of 3 mm to the canal, acut correction of tibial varus deformity was performed with tibial opened wedge osteotomy and fibular osteotomy performed through an incision of 1 cm, the procedure took approximately 2.5 hours. The conditions of the subjects were assessed during the first visit. The lengthening process was initiated after 5–7 days at a rate of 2 mm/day at the first week followed by 0.25 mm every 6h later. Simultaneously, the physical therapy was started to stretch and empower hip abductors, quadriceps femoris, calf muscles and Achilles tendon. Postoperatively when pain and swelling were reduced the patient was mobilized and after three weeks load bearing is allowed for both sides. The bone regeneration and deformity correction was usually assessed every two weeks by taking X-ray images. The rate of lengthening was adjusted to ensure the bone formation. The subjects were carefully investigated for possible complications. Once the desired length was achieved for both bones, one screw were inserted into the medullary nail at the proximal side of the femur, and the external fixators were both removed almost six weeks after the operation. The intramedullary nail supported the bone during the consolidation phase and allowed the removal of external fixator after the distraction phase of lengthening. Cefazolin (1 g/TDS) was administered during hospitalization, 7–10 days oral antibiotic was also prescribed. During the lengthening, in case of clinical diagnosis of infection (including pin-tract infection, osteomyelitis), appropriate antibiotic was administered. NSAIDs were prescribed for pain management as needed. The patient was followed up for six months so that their range of motion,

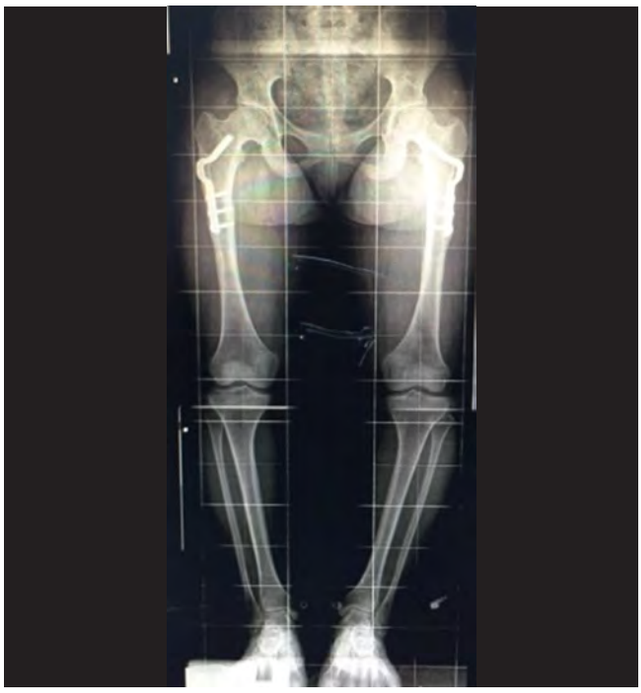

Figure 1. At the initial radiologic examination there were Harris Muller plates in both proksimal femurs from an operation that she had at two years of age.

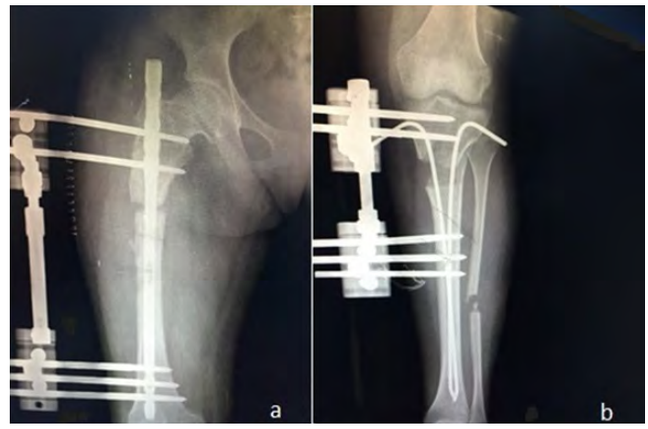

Figure 2. (a) After removing both plates right femur was leghthened with external fixator over an IM nail; (b) and the contralateral tibia with the same method correcting the tibial varus deformity. Because of the small calf of the tibia two of 3 mm IM elastic nails were used for tibial fixation.

level of pain and regenerated bone quality could be evaluated. Six mounts after the first operation we performed the same procedure for the left femur and the controlateral tibia. After one year follow up the patient was free of pain and had full range of motion for both lower extremities. The final results of the patient after removal of the lengthening device showed straight, re-aligned legs (Figure 3). Thus, feet are notably better positioned than prior to the correction, long-leg standing radiograph of the frontal plane mechanical axis showed optimal limb alignment and satisfactory length (5 cm for both) for the patient. There were no complications for the hole prosedure.

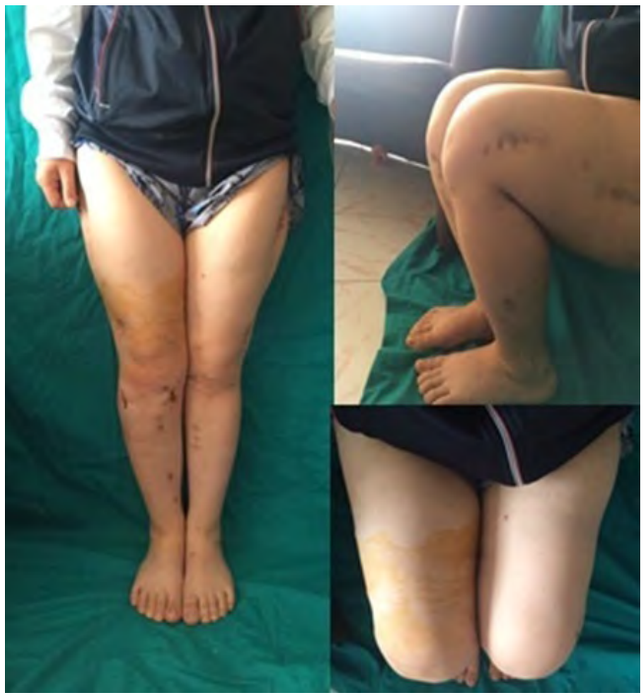

Figure 3. The final results of the patient after removal of the lengthening device showed straight, re-aligned legs with satisfactory length for the patient.

Discussion

Achondroplasia is the most common condition associated with disproportionate short stature. Surgical treatment to re-align the lower limbs in achondroplasia is generally indicated for cases who present either a severe, cosmetically unacceptable or clinically symptomatic limb deformity. Varus malalignment is generally more common than genu valga in this syndrome. Realignment can generally be achieved by gradual correction using external fixation devices [6]. Surgical correction is needed for tibial deformities higher than 10 degrees at coranal plane in adolescent with unexpected remodeling potential and adults [7]. Because of the the small size of the extremities especially in terms of patients with achondroplasia, external fixation itself can create discomfort. Because this application must be performed in multiple sessions, It is important for patients how much time they spend with external fixator. We performed simultaneous femoral and contralateral tibial lenghtening with correction of the tibial deformity. Thus we have shortened the duration of external fixation with fixator-assisted lengthening over an intramedullary nail. With earlier removal of the external fixator, the complication rates related to the pins, including pin tract infections and joint stiffness, are substantially diminished [8].

In conclusion fixator-assisted lengthening and deformity correction over an intramedullary nail in patients with achondroplasia is thought to be an appropriate and successful method with patients' high satisfaction.

Competing interests

The authors declare that they have no competing interests.

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Case Report
JCAM

A Rare Neurological Involvement in Sjögren's Syndrome: Abducens Nerve Palsy

Sjögren Sendromunda Nadir Görülen Nörolojik Bir Tutulum; Abdusens Sinir Felci

Abducens Nerve Palsy and Sjögren's Syndrome

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Özet

Sjögren sendromu (SS) ekzokrin bezlerin lenfositik infiltrasyonu ile karakterize otoimmun bir hastalıktır. Nörolojik tutulum hastaların yaklaşık dörttebirinde ortaya çıkmasına rağmen kranial sinir tutulumları çok daha nadiren görülmektedir. Burada kranial sinir tutulumu olan SS'lu bir vaka sunumu yapılmıştır.

Anahtar Kelimeler

Sjögren Sendromu; Kranial Nöropati; Abdusens Sinir Felci

Abstract

Sjögren's syndrome (SS) is an autoimmune disorder characterized by lymphocytic infiltration of exocrine organs. Although neurological involvement occurs in approximately one quarter of patients, involvement of cranial nerves is a relatively rare occurrence. Here a rare case of cranial neuropathy related to SS is reported.

Keywords

Sjögren's Syndrome; Cranial Neuropathy; Abducens Nerve Palsy

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Introduction

Sjögren's syndrome (SS) is an autoimmune disease characterized by chronic lymphocytic infiltration of exocrine glands. The prevalence of the disease ranges from 0.5 to 5%. SS can occur at all ages, with predominance in females between 40 and 50 years of age. The disease is nine times more commonly seen in females than in males. Primary Sjögren's syndrome (pSS) accounts for approximately 50% of all SS patients. Extraglandular symptoms such as articular, neurological, pulmonary, and hematological involvement are common over the course of the disease [1]. Neurological involvement often manifests as sensorial polyneuropathy. The involvement of cranial nerves is relatively rare [2]. In the published literature, isolated abducens nerve palsy has been reported in only 1 patient [3]. The current report presents a case of pSS with the involvement of cranial nerves.

Case Report

A 70-year-old female patient presented to an ophthalmology outpatient clinic complaining of double vision, headache, and nausea. The physical examination of the patient revealed limitation in lateral and medial gaze and ptosis. The patient was referred to the department of neurology with the suspicion of abducens nerve palsy (Figure 1). The patient's medical history

Figure 1. Abducens nerve palsy in the patient

was not remarkable, with the exception of preexisting hypertension and cranial diffusion on magnetic resonance imaging (MRI). MR venography was performed to exclude intracranial hypertension, sinus vein thrombosis, and intracranial space occupying lesions. Carotid-vertebral artery computed tomography (CT) angiography did not show any intracranial aneurysm, and Doppler ultrasonography of the temporal arteries revealed normal findings. Orbital MRI was also normal. The examination of the cerebrospinal fluid (CSF) did not show pathological findings (angiotensin converting enzyme (ACE), adenosine deaminase, Lyme serology, tuberculosis culture, or brucella). To exclude myasthenia gravis repetition, electromyoneurography (EMNG) was performed and revealed normal findings. The patient also tested negative for acetylcholine antibodies. Peripheral blood test showed the following: WBC: 12.9 x103 µL, Hb: 16.5 gr/dl, PLT: 220.000 x103 µL, ESR: 40 mm/h, C-reactive protein: 6.7 mg/dl, C3: 114 (90-180), C4: 14 (10-40), IgG: 753 mg/dl (700-1600), anti-HCV (-), rheumatoid factor: 167 IU/ml (0-15); anti-cyclic citrullinated peptide antibody (-) and urine analysis showed normal findings. The assays performed due to the suspicion of connective tissue disease and vasculitis showed 4+ antinuclear antibody (ANA) with centromere pattern. In light of these findings, a rheumatologist was consulted. Extractable nuclear antigens and antineutrophil cytoplasmic antibodies were negative. SS-A antibody was positive. Echocardiography showed normal pulmonary artery pressure and chest x-ray showed normal findings. There was no clinical finding suggestive of scleroderma. Schirmer's test was performed due to the coexistence of dry mouth and eye complaints with joint pain. The test results were 3 mm in the right eye and 2 mm in the left eye. Because the patient was found to have dry eye syndrome, asalivary gland biopsy was performed; it revealed diffuse lymphocytic infiltration (grade 4). The patient was therefore diagnosed with pSS with the involvement of cranial nerves and was placed on a therapy with methylprednisolone 1 mg/kg/day, azathioprine 100 mg/day, and hydroxychloroquine 400 mg/day. Double vision, ptosis, and limitation in lateral and medial gaze showed almost complete recovery.

Discussion

Sjögren's syndrome is known to cause neurological involvement in approximately 25-30% of cases [2]. In the cohort of Jamilloux et al., neurological involvement occurred in 95 (22%) out of 420 patients with SS. SS may affect both the peripheral nervous system (PNS) and central nervous systems (CNS) (Table 1). The pathogenesis of neurological involvement has not been

Table 1. Neurological involvement in Primary Sjögren's Syndrome

PNS	CSS
Sensory neuropathy	Focal manifestations
- Small fiber neuropathy	Aseptic meningoencephalitis
- Sensory ataxic neuropathy	Myelopathy
Sensorimotor polyneuropathy	Headache
Mononeuritis multiplex	Cognitive disorders
Demyelinating polyradiculoneuropathy	Mood disorders
Cranial nevre involment	Seizure
Autonomic neuropathy	Pyramidal signs
	Tranverse myelitis

PNS: Peripheral nervous system, CSS: Central nervous system

fully elucidated. Vasculitic lymphocytic infiltration of vasa nervorum has been the most commonly implicated mechanism in the involvement of the PNS regardless of the presence of necrosis [4]. Two hypotheses have been set forth to explain CNS involvement. The first hypothesis suggests direct infiltration of the CNS by mononuclear cells, whereas the second hypothesis suggests vascular involvement. Vasculopathy has been considered to be associated with the presence of anti-Ro and antineural antibodies [5].

Sensorial polyneuropathy is the most common type of PNS involvement, occurring in approximately 20% of cases [2]. The patients with Sjögren's often present with numbness, burning, and pain originating in distal parts of the extremities. Mononeuropathy is relatively rare in SS. Mononeuropathy encompasses mononeuritis multiplex, cranial neuropathy, and trap neuropathies. Cranial neuropathies show symptoms depending on the cranial nerve involved. Among cranial nerves, the trigeminal nerve and optic nerves are the most commonly involved [3].

Visual changes can be quite profound in Sjögren's. The most worrisome is inflammation around or behind the eye, called optic neuritis or retrobulbar neuritis. This is treatable, but there is potential risk of loss of vision. This type of inflammation can also be seen in a variety of other neurologic conditions. There

is a similarity that sometimes bedevils us between multiple sclerosis and Sjögren's involvement in the nervous system. The aspect that makes differentiating between the diseases most difficult is involvement of the eye in optic neuritis.

The treatment usually involves the use of immunosuppressive medications such as corticosteroids, cyclophosphamide, and azathioprine. Intravenous immunoglobulin therapy is used in refractory cases. However, intravenous immunoglobulin therapy has some disadvantages due to indefinite dosage, the duration of therapy, and high treatment costs. There is a need for randomized controlled studies in order to establish the effectiveness of this therapy [6]. Rituximab, an anti-CD 20 antibody, offers promising results; however, further comprehensive studies are required in order to confirm its efficiency [7].

In conclusion, extraglandular organs can be involved in patients with Sjögren's syndrome. In clinical practice, it should be considered that neurological involvement occurs in one quarter of SS cases. In fact, neurological complaints may be the first sign of SS. We emphasize that connective tissue disorders should be considered in patients presenting with similar complaints; it is prudent to evaluate these patients for the possibility of pSS.

Competing interests

The authors declare that they have no competing interests.

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Simultaneous Radial Lengthening and Ulnar Shortening for a Delayed Presentation of Radius Distal Physeal Arrest: A Case Report

Distal Radius Fizyal Arreste Bağlı Eş Zamanlı Radius Uzatma ve Ulna Kısaltma: Vaka Sunumu

Simultaneous Radius Lengthening and Ulnar Shortening

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Özet

Distal radius kırıkları çocuklar ve yaşlılarda sık görülen el bileği yaralanmalarıdır (% 18% 25). Distal radius kırığı sonrası büyüme plağının etkilenmesi ve fizyal arrest gelişmesi ise daha nadirdir. Fizyal arrest sonrası gelişen ana deformite radius kısalığı ve bununla ilişkili olarak ulna aşırı büyümesidir, el bileği ağrısı ve fonksiyonel kısıtlılık ise ana yakınmalardır. Bu yazıda travma sonrası distal radius fizyal arrest gelişen adölosan hastanın deformitesine yönelik uygulanan eş zamanlı radyal uzatma ve ulnar kısalma cerrahi prosedürünün sunulması amaçlanmıştır.

Anahtar Kelimeler

Radiyal Uzatma; Ulna Kısaltma; Eşzamanlı Osteotomi; Fizyal Arrest; Pozitif Ulnar Varyans

Abstract

Distal radius fractures are common injuries in both children and in the elderly (25%; 18%). Distal radius physeal fractures have a high incidence, but physeal growth arrest occurs at a low rate. As a main deformity, radial shortening occurs with relative ulnar overgrowth leading to significant complaints of pain and functional limitations after distal radial growth arrest. In this paper we aim to report on the restoration of the wrist mechanics attained by performing a surgical technique of simultaneous radial lengthening and ulnar shortening procedures in an adolescent with a significant ulnar overgrowth deformity due to a posttraumatic growth arrest of distal radius.

Keywords

Physeal Arrest; Positive Ulnar Variance; Treatment; Radial Lengthening; Ulnar Shortening; Simultaneous Osteotomy

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Introduction

Distal radius fractures are common injuries in both children and the elderly (25%; 18%) [1]. Although physeal fractures of the distal radius have a high incidence, physeal growth arrest occurs at a low rate (1%-7%) [1]. As a main deformity of distal radial growth arrest, radial shortening occurs with relative ulnar overgrowth leading to significant complaints of pain and functional limitations. The deformity of disturbed radio-ulnar variance is mostly associated with triangular fibrocartilage complex (TFCC) lesions and distal radio-ulnar joint (DRUJ) instability. If the dome of the distal ulna is more distal than the ulnar corner of the distal radius it is called 'positive ulnar variance' [2]. Positive ulnar variance can be the reason for the wrist pain, abnormal wrist mechanics, and limited ulnar deviation and rotation of the forearm. To reestablish a neutral radio-ulnar variance and to achieve the best possible functional results, as many authors described, ulnar shortening or radial lengthening osteotomies can be performed for this type of injury. In the literature, in a symptomatic patient with positive ulnar variance, the largest ulnar shortening performed was 15 mm in length [3]. Taylor spatial frames and external fixators are more commonly used for the correction of radial deformity and length. Ulnar shortening osteotomy is considered the standard procedure for correcting positive ulnar variance [4]. In the literature there are some studies comparing the two techniques, but our search indicates that there is no study about simultaneously performing radial and ulnar osteotomies for radial growth arrest deformity. We think that radial lengthening osteotomy can be added to ulnar shortening for more severe deformities.

In this paper we report the treatment of both acute ulnar shortening and gradual radial lengthening for the ulnar overgrowth deformity in a case of an adolescent with a significant growth arrest of the distal radius and with significantly limited ulnar deviation.

Case Report

An adolescent 16-year-old male presented to our outpatient clinic with complaints of ulnar-sided wrist pain, wrist deformity, and difficulty with movements, especially opening doors with his dominant right hand. The patient's stature was normal for his age. Reporting no family history or previous surgery of the affected extremity, he gave the history of previous treatment of distal radius fracture with closed reduction and casting, 7 years before at the same extremity. He first noticed the deformity 2 years after the fracture and the deformity increased with time. When he experienced difficulties with daily activities and increased pain with wrist movements, he came to our outpatient clinic. At presentation, there was a prominence of the distal ulna and radial deviation deformity at his right dominant wrist. At the wrist joint flexion, extension and pronation was normal but supination was limited at nearly 30°. There was a severe pain with movements, especially of ulnar deviation and supination. At the radiography, XR imaging showed nearly a 17 mm positive ulnar variance (Figure 1). The contralateral wrist had no physical or radiologic abnormalities. We considered undertaking an acute ulnar shortening osteotomy together with gradual radial lengthening osteotomy because of the severe ulnar overgrowth deformity. We notified the patient and his family

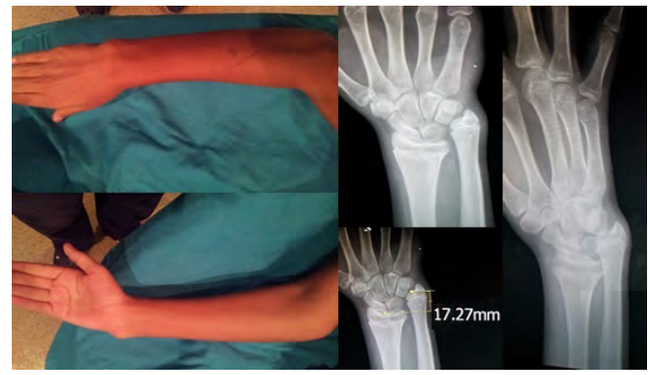

Figure 1. Preoperative photograph and XR imaging showing nearly a 17 mm positive ulnar variance

about the procedure, the possible complications, as well as the duration of the process. Informed consent was obtained from the family. Under general anaesthesia with a direct approach to distal ulna, making an oblique osteotomy, first we performed 10 mm shortening. For fixation, a 6-hole dynamic compression plate was used (Figure 2). Then with a dorsal approach to distal radius, a metaphyseal osteotomy was performed. Then the osteotomy side was fixed with a monolateral external fixator, inserting both two 3.3mm schantz screws proximally and distally (Figure 2). To control the rotation, both osteotomy sides were marked. After the operation we set the forearm in a spina cast for 2 weeks. After removing the cast, radial lengthening process was initiated at a rate of 1 mm/day. Simultaneously, physical therapy was started to regain wrist movements. After 10 days we stopped lengthening. The monolateral external fixator supported the bone for 3 more weeks during the consolidation phase and allowed for the removal of the external fixator 6 weeks following the operation. The bone regeneration and deformity correction were usually assessed every two weeks by taking X-ray images. The rate of lengthening was adjusted to ensure the bone formation. Cefazolin (1 g/TDS) was adminis-

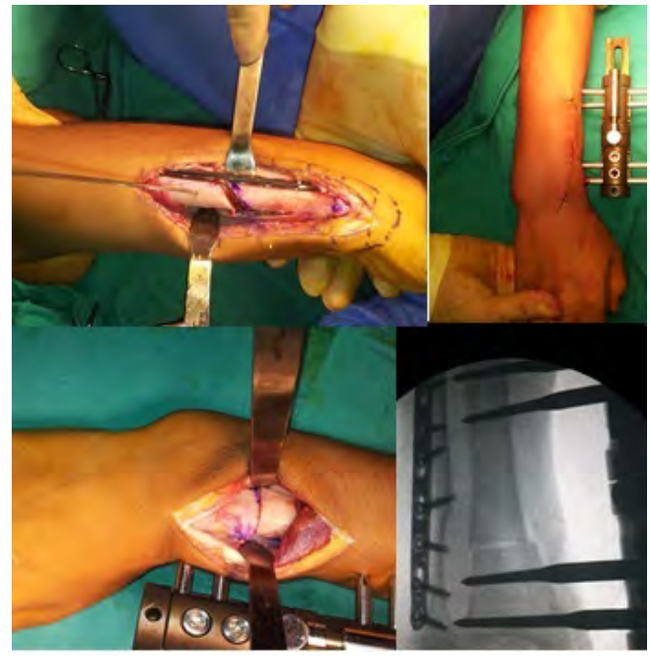

Figure 2. Intraoperative photograph showing distal ulna oblic osteotomy and fixation with a 6-hole dynamic compression plate. Distal radius metaphyseal osteotomy fixation with monolateral external fixator inserting both two 3.3mm schantz screws proximally and distally.

tered during hospitalization and 7–10 days oral antibiotic was also prescribed. During the lengthening, in case of a clinical diagnosis of infection (including pin-tract infection, osteomyelitis), appropriate antibiotic was administered. Nonsteroidal anti-inflammatory drugs were prescribed for pain management as needed. The patient was followed up for six months so that his range of motion, level of pain, and regenerated bone quality could be evaluated. Three years after the surgery, the patient had no ulnar prominence or pain with movements and had significant improvement of forearm supination. The X-ray imaging showed that the osteotomy sides were both healed with a neutral radio-ulnar variance (Figure 3). There were no complications during the follow-up period.

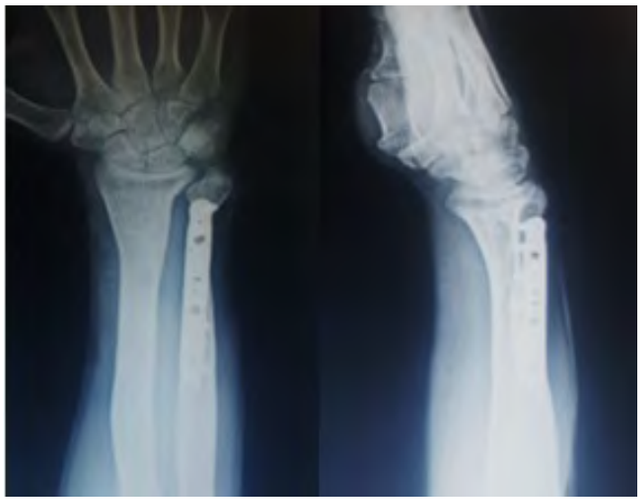

Figure 3. The final control X-Ray imaging showing that the osteotomy sides were both healed with a neutral radio-ulnar variance.

Discussion

Although physeal fractures of the distal radius are common injuries in children, physeal growth arrest occurs at a low rate (1%-7%) [1]. In the literature there are several studies including case reports and case series regarding the surgical management of distal radial growth arrest resulting in ulnar overgrowth deformities [3,5]. There are different techniques such as ulnar shortening and radial lengthening osteotomies to reestablish a neutral radio-ulnar variance for this type of deformities. Ulnar shortening osteotomy is considered the standard procedure for correcting positive ulnar variance [4]. External fixators, radial volar plates with grafting, or Taylor spatial frames are more commonly used for the correction of radial deformity and length [5,6]. Although different methods were presented for the surgical treatment of positive ulnar variance deformity, in severe deformities acute correction can sometimes be difficult to achieve, as radial lengthening osteotomy requires a large amount of strut bone graft, and ulnar shortening osteotomy of over 1 cm is difficult owing to soft tissue contracture [7]. Supporting this idea, Bowers [7] pointed out that ulnar shortening of more than 5–6 mm presents several problems with alignment and matching of bone ends because of soft tissue pressure on the construct. Miura et al. [8] also reported that distal radio-ulnar joint acute reduction can cause excessive joint loading and joint pain, stiffness, or instability. As an alternative Gong et al. [6] have reported satisfactory results after the gradual correction of distal radial malunion or physeal arrest using distraction osteogenesis. In the literature, in a symptomatic patient with positive ulnar variance, the largest ulnar shortening performed was 15 mm in length [3]. We here report a case of a delayed presentation of radius distal physeal arrest due to a prior fracture in an adolescent with nearly 17 mm positive ulnar variance. Because of the limitation of ulnar shortening, we considered making another osteotomy simultaneously to the distal radius to gradually lengthen the radius with a monolateral external fixator until we reestablished the neutral radio-ulnar variance. We performed ulnar oblique osteotomy because of the faster healing rate and lower nonunion rate when compared to the tranverse osteotomy. Using gradual lengthening of the radius, we allowed the distal radio-ulnar joint sufficient time for soft tissue adaptation. At the final follow up rebalancing the wrist, we had satisfactory results as improved range of motion, improved function, and decreased wrist pain were achieved. Also we had no complications such as pin-tract or wound infection, delayed union, or nonunion of the osteotomy side. We think while the ulnar shortening osteotomy is a suitable procedure for distal radial growth arrest in patients with little or no growth potential, radial lengthening osteotomy can be added for more severe deformities, affording a good outcome and restoration of the radio-ulnar variance.

Conclusion

In symptomatic adolescents, surgery for posttraumatic distal radial growth arrest can reduce the level of pain and loss of motion. To reestablish a neutral radio-ulnar variance and to achieve the best possible functional results for more severe positive ulnar variance deformities, ulnar shortening and radial legthening osteotomies can be performed simultaneously.

Competing interests

None of the authors has any conflict of interest to declare.

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Acquired Hypothyroidism in an Adolescent Associated with Radioactive Iodine-131 Therapy of the Mother

Radyoaktif İyot-131 Tedavisi Alan Annenin Adolesan Çocuğunda Gelişen Hipotiroidi

Hypothyroidism and I131 Treatment

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Özet

Çocukların radyoaktif iyota maruz kalması sonucu hipotiroidi ve tiroid kanserleri gelişebilir. Bu yüzden özellikle küçük çocuk ve gebelerin bu tedaviyi alanlardan bir süre uzak tutulması gerekir. Literatürde bu tedaviyi alanlarda kendilerinin tedavi alması sonucu iki olgu ve annesinin aldığı tedaviden etkilenerek bir olgu olmak üzere toplam sadece üç tiroid kanseri geliştiren adolesan olgu mevcuttur. Bu olgu sunumunda, radyoaktif iyot tedavisi alan bir anneyle aynı ortamda yaşayan ve annesinin bu tedavisine bağlı olarak hipotiroidi gelişen 11 yaşındaki kız olgu sunulup, literatür bilgileri ışığı altında tartışılması planlandı.

Anahtar Kelimeler

Çocuk; Hipotiroidi; Radyoaktif İyot

Abstract

Hypothyroidism or thyroid cancer may develop in children as a result of exposure to radioactive iodine. Therefore, young children and pregnant women in particular should not be in close physical proximity to patients receiving this therapy. We found only three cases of adolescents developing thyroid cancer in the literature. Two were affected by radioactive iodine treatment of themselves and one was affected by treatment of the mother. Here we report on an 11-year-old girl living in the same environment as her mother, who was receiving radioactive iodine therapy. The girl developed transient hypothyroidism in association with her mother's treatment. We discuss the case in light of the literature.

Keywords

Children; Hypothyroidism; Radioactive Iodine

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Introduction

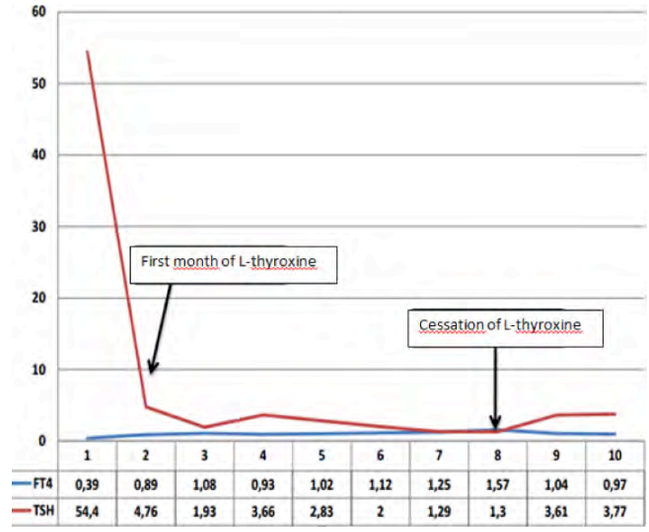
Radioactive iodine (131I) therapy is an effective, economical, and simple method, used widely, particularly to treat Graves' disease and thyroid gland malignities in adults. However, due to insufficient evidence regarding its reliability, it is still not widely used in children. Hypothyroidism, autoimmune thyroiditis, and thyroid neoplasms may develop as the result of accidental exposure of children to radioactive iodine [1]. All people, and particularly young children and pregnant women, are advised to avoid being in proximity to patients receiving this treatment. Cases of exposure in the prenatal period and the resultant development of congenital hypothyroidism and thyroid nodules have been widely reported in the literature [2,3]. Thyroid cancer has been reported in two adolescents receiving radioactive MIBG therapy, and there has been only one publication reporting papillary thyroid carcinoma developing as a result of 131I therapy administered to the mother [4,5]. Here we report a case of hypothyroidism in the 11-year-old child of a mother receiving 131I therapy due to thyroid cancer.

Case Report

An 11-year-old girl presented to our clinic with symptoms of lethargy, fatiguing easily, and swelling in the neck region over the previous 1.5 months. Her mother had been operated on due to papillary thyroid carcinoma approximately 2 months previously, after which she had received 131I therapy. The mother's two other children had been removed from the home due to the radioactive therapy she received, but our patient had remained with the mother. At physical examination, the patient's weight was 26 kg (3%) and height was 132.5 cm (10%). Head and neck examination revealed stage 1 thyromegaly. No tachycardia was observed, and arterial blood pressure values were normal for her age (110/65 mmHg). Laboratory examination revealed fT4: 0.39 ng/dL (N= 0.61-1.12 ng/dL) and TSH: 54.4 uIU/mL (N=0.34-4.2 uIU/mL), while anti-TPO and anti-thyroglobulin were negative. At thyroid ultrasonography, the left and right thyroid lobes were large for her age, while the thyroid parenchyma was homogeneous. No lymph nodes were pathological in size. The patient was started on L-thyroxin 2 mcg/kg/day due to hypothyroidism. At thyroid function follow-up one month later, the patient was euthyroid while the right and left thyroid lobes had decreased to an age-appropriate size (Figure 1). The case was monitored as euthyroid under low-dose L-thyroxin therapy for 2 years. Growth rate, weight gain, and pubertal development were within physiological limits throughout monitoring. Treatment was concluded at the end of 2 years.

Discussion

Immune system-related causes and iodine deficiency are the most important factors in the etiology of childhood hypothyroidism. In our patient who presented with hypothyroidism, the medical history revealed no etiological factor, other than the mother's having received 131I therapy due to papillary cancer. The fact that the child's symptoms developed 15 days after the mother's receipt of this treatment, and the exclusion of other causes, suggested that exposure to radioactive 131I in the child, who had not been removed from the mother's presence during her 131I therapy, was involved as the agent in the disease.



	1	2	3	4	5	6	7	8	9	10
FT4	0,39	0,89	1,08	0,93	1,02	1,12	1,25	1,57	1,04	0,97
TSH	54,4	4,76	1,93	3,66	2,83	2	1,29	1,3	3,61	3,77

Figure 1. The course of thyroid function

Radioactive iodine therapy is an effective, economical, and simple method that has been used to treat hyperthyroidism and thyroid neoplasms in adults for approximately 50 years. The radiation damages thyroid follicle cells and prevents hormone synthesis. This method is contraindicated in pregnancy and lactation. It is not popular with pediatricians because tissues in early adulthood and childhood are more sensitive to radiation. Individuals receiving radioactive iodine therapy must avoid pregnant women and children for at least 1 week for safety reasons, must use separate eating and cleaning implements than other members of the family, must keep a distance of at least 60 cm between themselves and others, and must sleep alone. The relationship between radioactive iodine and malignity has not been proven. However, data from nuclear accidents have shown an increase in cases of non-immune hypothyroidism and thyroid cancers among exposed individuals [1,6,7]. Thyroid cancer was reported to develop in two adolescents receiving 131I MIBG therapy for neuroblastoma [4]. Cases of congenital hypothyroidism and thyroid cancer have been reported to develop following intrauterine exposure [2,3]. However, we encountered only one case of thyroid cancer developing in a child in close proximity to an individual receiving treatment [5]. We encountered no reports of transient hypothyroidism as was seen in our case. We attributed thyroid follicle cell injury and transient hypothyroidism in our patient to the radioactive iodine therapy received by the mother. The laboratory findings in our case showed that hypothyroidism developed due to non-immune causes; the condition was temporary since it resolved completely with monitoring and treatment. Although there are previous cases in the literature of congenital hypothyroidism developing as an effect of radioactive iodine administered to the mother in the intrauterine period, to the best of our knowledge, ours is the first case of hypothyroidism developing in an adolescent child exposed to radioactivity via the mother. We think that this report will make a significant contribution to the literature, and that greater care should be taken over safety precautions following radioactive iodine therapy.

Competing interests

The authors declare that they have no competing interests.

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Postpartum Osteoporosis and Thoracic Vertebral Fracture in a Patient Treated with Heparin During Pregnancy

Gebelik Sırasında Heparin Tedavisi Alan Bir Hastada Postpartum Osteoporoz ve Torakal Vertebra Kırığı

Postpartum Osteoporosis and Heparin

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Özet

Postpartum Osteoporoz (PPO) gebelik ile ilişkili, nadir görülen bir osteoporoz tablosudur. Biz, doğumundan 1 hafta sonra şiddetli bel ağrısı şikayetiyle başvuran otuz beş yaşında bir kadın hasta sunduk. Hasta, protein C eksikliği nedeniyle gebeliği boyunca düşük molekül ağırlıklı heparin (DMAH) 40 mg/gün tedavisi almış. Vücut kitle indeksi (BMI) 19,8 ve gebeliği boyunca sadece 8 kg almış. Manyetik rezonans görüntülemede (MRG) torakal 11'de bir fraktür saptandı. Dual-enerji Xray absorpsiyometri (DEXA) Lomber1-4 vertebralarda T skoru= - 4,9 ve Z skoru= -4,8 ölçüldü. Bu bulgular, PPO'un postpartum hastalarda şiddetli bel ağrısı nedenlerinden biri olabileceğini düşündürmektedir. Özellikle düşük BMI'li ve gebelik sırasında DMAH kullanan hastalarda, PPO riskinin daha yüksek olduğunu düşünüyoruz.

Anahtar Kelimeler

Postpartum Osteoporoz; Heparin; Kemik Mineral Dansite; Fraktür

Abstract

Postpartum osteoporosis (PPO) is a rare form of osteoporosis related to pregnancy. We report the case of a 35-year-old woman who consulted for severe low-back pain one week after her delivery. This woman had a personal history of protein C deficiency and was treated with low-molecular-weight heparin (LMWH) 40 mg/day during her pregnancy. Her body mass index was 19.8 and she had only gained 8 kg during pregnancy. Magnetic resonance imaging (MRI) revealed a fracture of thoracic 11. Dual-energy X-ray absorptiometry (DEXA) measured T score = - 4,9 and Z score = -4,8 in Lumbar 1-4 vertebrae. These findings suggest that PPO may be one of the causes of severe back pain in postpartum patients. We think that PPO risk is higher in those patients with low BMI who were treated with LMWH during pregnancy.

Keywords

Postpartum Osteoporosis; Heparin; Bone Mineral Density; Fracture

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Introduction

PPO presents symptomatic vertebral compression fractures causing severe pain in the thoracic or lumbar area. It is not clear why some people are prone to develop PPO; however, hypotheses such as genetic susceptibility, an inherited defect in collagen synthesis, and an exacerbated response to physiological hormonal changes have been suggested [1].

Long-term unfractionated heparin (UFH) use is associated with osteoporosis in both non-pregnant and pregnant patients. Although the exact mechanism is unknown, possible mechanisms are decreased osteoblastic and increased osteoclastic activity caused by direct effect of heparin, vitamin D deficiency, and decreased serum ionized calcium concentrations resulting in increased PTH stimulated bone resorption [2]. In this case, we report severe PPO and a thoracic vertebral compression fracture diagnosed in the postpartum period, in a patient treated with LMWH throughout her pregnancy.

Case Report

We report the case of a 35-year-old woman who consulted for low-back pain one week after her first delivery. The second day after birth, she reported a severe back pain while she was getting up from the bed. In her history, she had protein C deficiency and had been treated with LMWH 40 mg/day during pregnancy for prophylaxis of thromboembolism. Her first pregnancy was terminated due to intrauterine exitus of fetus. The second pregnancy was completed as healthy. Menses of the patient occurred only 2 times in 16 months during this time period. Because of the risk of abortion, she had been immobilized during pregnancy. Her weight was 54 kg before pregnancy and her height was 165 cm, revealing a body mass index (BMI) of 19.8 kg/m2; she gained only 8 kg during pregnancy. A nutritional questionnaire showed that her daily calcium intake was between 800 and 1000 mg. She was healthy and had regular menses before pregnancy. There was no history of the use of drugs, such as corticosteroids or thyroid hormones that could affect bone metabolism, and no personal or family history of osteoporosis or fractures. She was a non-smoker. On physical examination, the patient had kyphosis and pain and tenderness on the thoraco-lumbar vertebrae with palpation. Her serum calcium, phosphorus, albumin, prolactin, and cortisol levels, thyroid hormones, and 25-hydroxyvitamine D were normal (Table 1). Magnetic resonance imaging (MRI) revealed height loss in thoracic 11, which was surrounded by a diffuse low bone marrow signal (Figure 1). Bone mineral density (BMD) was measured by using dual-energy X-Ray absorptiometry (DEXA), and very low T and Z-scores were determined in lumbar vertebrae (Table 2). According to these findings, she was diagnosed with severe postpartum osteoporosis (PPO). After analgesia and 1 month of home relative bed rest, lactation was stopped and Alendronate 70 mg/week, calcium 1000 mg/day, and vitamin D 880 U/day were prescribed. Her pain was decreased at the third month.

Table 1. The laboratory findings of the patient.

Calcium (8,8-10,6 mg/dl)	9,7
Phosphorus (2,5-4,5 mg/dl)	4,1
Albumin (3,5-5,2 mg/dl)	4,4
Parathyroid hormone (15-68,3 pg/ml)	70,8
25(OH)D3 (10-60 ng/ml)	32,6
Thyroid stimulating hormone (0,35-4,94 mIU/ml)	0,53±0,5
Free triiodothyronine (1,71-3,71pg/ml)	3,11
Free thyroxine (0,9-1,7 ng/dl)	0,8
Cortisol (6,2-19,4 mcg/dl)	9,8
Prolactin (5,18-26,53 ng/ml)	36,9

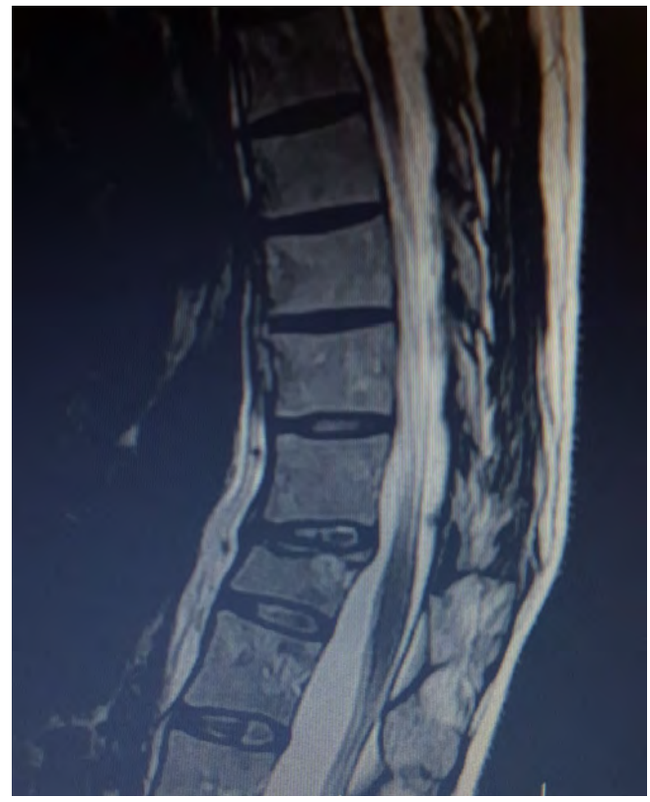

Figure 1. Vertebral compression fracture in magnetic resonance imaging in thoracic 11.

Table 2. Dual-energy X-ray absorptiometry results of the patient.

Lomber 1	
BMD (gr/cm2)	0,489
T	-4,0
Z	-3,9
Lomber 2	
BMD (gr/cm2)	0,530
T	-4,5
Z	-4,4
Lomber 3	
BMD (gr/cm2)	0,486
T	-5,4
Z	-5,3
Lomber 4	
BMD (gr/cm2)	0,512
T	-5,5
Z	-5,4
Lomber 1-4	
BMD (gr/cm2)	0,504
T	-4,9
Z	-4,8
Femur neck	
BMD (gr/cm2)	0,491
T	-3,2
Z	-3,0

Discussion

In this case report, we presented a vertebral compression fracture and severe PPO in a woman who used LMWH throughout her pregnancy. According to X-ray and MRI findings, we found a fracture in thoracic 11. Dual-energy X-ray absorptiometry (DEXA) showed very low T and Z-scores in lumbar vertebrae and the femur neck.

Postpartum osteoporosis (PPO) is a rare disease presenting with back pain and, at times, multiple vertebral fractures during late pregnancy or during the early postpartum period.

Although etiology and pathogenesis have not been clarified yet, possible mechanisms include: increased bone turnover to meet the calcium requirements of the fetus, increased serum parathyroid hormone (PTH), relative hypoestrogenemia and high prolactin levels, and presence of a genetic background [3]. During pregnancy and lactation, BMD was found to be decreased in some studies [4, 5], while it did not change in others [6, 7]. In the literature, various risk factors were identified for PPO. Amenorrhea and oral contraceptive treatment, suppressive levothyroxine treatment, osteogenesis imperfecta, low body mass index and weight loss, physical inactivity, and corticosteroid therapy are some of these risk factors [8-12]. However, genetic factors also play a major role in the pathogenesis of osteoporosis. Genetic studies indicate that osteoporosis is a polygenetic disorder resulting from the interaction between common polymorphic alleles and multiple environmental factors [13]. The risk of developing osteoporosis is believed to be substantially lower when LMWHs are preferred for prophylaxis or treatment of thromboembolism [14].

Long-term heparin therapy is associated with both an increase in osteoclast activity and a suppression of osteoblast function [15]. Heparin-induced osteoporosis has been reported in patients receiving heparin at daily doses of 15,000 units or more for at least 6 months [16]. LMWH suppresses osteoblast function, but may not increase osteoclastic activity, and this may explain why there appears to be a lower risk of osteoporosis with LMWH [17]. On the other hand, experimental animal studies have shown that unfractionated heparin (UFH) and LMWH exert similar unfavorable effects on histomorphometric parameters of cancellous and cortical rat bone [18, 19], and so the notion that LMWH is less likely to lead to osteoporosis and bone fracture remains somewhat controversial.

In rats, after the administration of enoxaparin, the ratio of bone mineral content to bone mass decreased, bone formation was inhibited, and bone resorption was intensified [18]. In a study evaluating patients receiving enoxaparin for 3–24 months, a modest but progressive decrease in BMD was observed, and the authors advised performing densitometry before starting long-term anticoagulation, especially in patients with concomitant risk factors for osteoporosis.

According to our knowledge, in the literature, there are few patients with PPO possibly associated with LMWH. Goëb et al. reported low-back and right-buttock pain and fracture of the right sacral ala in a 19-year-old woman treated daily with LMWH during her pregnancy [20]. Since she had no potential clinical risk factors for osteoporosis, the causal effect of LMWH was suggested. In another report, PPO and vertebral fractures were observed in a 40-year-old lactating woman who received LMWH in the final 2 trimesters of pregnancy [21]. Ozdemir et al. showed PPO and vertebral fractures in two patients treated with enoxaparin during pregnancy. The first patient's age was 34, and the second was 36 years old. The two patients had multiple vertebral fractures and decreased BMD and body mass indexes of 21.9 kg/m2 and 22.4 kg/m2 respectively [22]. Because of the risk of abortion, our patient had been immobilized during pregnancy. She had a low BMI throughout her adult life. To our knowledge, the risk factors for our patient were weight loss, physical inactivity, and treatment with LMWH during pregnancy.

In conclusion, PPO should be included in the differential diagnosis of severe back pain in pregnant and postpartum patients. Postpartum women presenting with the sudden onset of low-back pain, along with a diagnosis of a herniated disc or a mechanical low-back pain, PPO should be considered in the differential diagnosis. Especially in patients with low BMI using LMWH during pregnancy, we think that the risk is higher for PPO. In addition to the use of LMWH, immobilizing lowers the patient's risk of abortion, but we believe that it further increases the risk for PPO. In these patients, the diagnosis should be confirmed by MRI. Early radiographs are often inconclusive but are necessary. However, the diagnosis of PPO may be delayed. MRI may be a better imaging method for breastfeeding mothers.

Competing interests

The authors declare that they have no competing interests.

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Sepsis-Associated Encephalopathy in a Child with the Torsion of Meckel's Diverticulum

Meckel Divertikülü Torsiyonu Olan Bir Çocukta Sepsis İlişkili Ensefalopati

Sepsis-Associated Encephalopathy

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Özet

Sepsis-ilişkili ensefalopati enfeksiyona sistemik yanıta ikincil gelişen yaygın bir beyin fonksiyon bozukluğudur ve yüksek mortalite oranı ile ilişkilidir. Biz 4 yaşındaki sepsis-ilişkili ensefalopati görülen bir erkek hastayı sunduk. Hasta karın ağrısı ve kusma ile başvurdu. Yatışının ikinci gününde, dikkat bozukluğu, konfüzyon ve deliryumun eşlik ettiği bilinç bozukluğu geliştirdi. Rutin laboratuar testleri, beyin manyetik rezonans görüntüleme ve beyin omurilik sıvısı incelemesi normaldi. Elektroensefalografide sağ hemisferde epileptiform deşarjların eşlik ettiği yüksek voltajlı yavaş dalga aktivitesi izlendi. Hasta acilen ameliyata alındı ve tortiöz, gangrenöz Meckel divertikülü ve komşuluğundaki ince bağırsağa uzanan iskemi alanı görüldü ve bu alanlar çıkarıldı. Hastanın üçüncü gün bilinci normale döndü ve izlemde sekelsiz olarak taburcu edildi. Sepsis-ilişkili ensefalopati hastalarında kötü prognozun üstesinden gelmek için, belirtilerinin erken tanınması ve altta yatan nedenin uygun tedavisi çok önemlidir.

Anahtar Kelimeler

Sepsis İlişkili Ensefalopati; Meckel Divertikülü; Çocuk

Abstract

Sepsis-associated encephalopathy (SEA) is a diffuse brain dysfunction secondary to the systemic response to infection and is associated with high mortality rate. We report a 4-year-old boy with SEA. He presented with abdominal pain and vomiting. On the second day of admission, he developed consciousness disturbance with impaired attention, confusion and delirium. Routine laboratory tests, brain magnetic resonance imaging and cerebrospinal fluid examination were normal. Electroencephalography (EEG) showed high-voltage slow wave activity on the right hemisphere with epileptiform discharge. He immediately underwent surgery and a torsed, gangrenous Meckel's diverticulum with extension of ischemia to adjacent small bowel was seen and resected. His consciousness had become normal by the third day and he was discharged without any sequela. To overcome a poor prognosis in patients with SEA, the early recognition of the symptoms of SEA and also appropriate treatment of the underlying cause are essential.

Keywords

Sepsis-Associated Encephalopathy; Meckel's Diveticulum; Children

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Introduction

Sepsis-associated encephalopathy (SEA) is the most common form of encephalopathy in intensive care unit and has been associated with a higher mortality rate [1]. An encephalopathy with variable severity is found to occur in 9-71% of septic adult patients [2]. The frequency of SEA or any other neurologic disability has not been demonstrated in children with sepsis [3].

Meckel diverticulum is the most common congenital abnormality of the gastrointestinal tract, resulting from the failure of obliteration of omphalomesenteric duct. The majority of patients with Meckel diverticulum are asymptomatic. Only 4-6% of Meckel's diverticulum is symptomatic [4,5]. Hemorrage, intestinal obstruction and inflammation are the most common complications related with Meckel's diverticulum [4]. Axial torsion and gangrene of Meckel's diverticulum are the rarest complications, especially in children [6].

We report a 6 years old boy with SEA complicating the torsion of Meckel's diverticulum.

Case Report

A 6-year-old boy presented to the emergency department with abdominal pain and vomitting. On admission, the clinical examination revealed minimal periumblical pain. The leucocyte count was elevated at 18000/mm3 with 87 percent segmented neutrophils. Also C-reactive protein was 69.1 mg/dl (normal < 5). The ultrasound examination was normal. On the second day of admission, he developed consciousness disturbance with impaired attention, confusion and delirium. His psychomotor development had been normal and there was no family history of neurological disorders. On neurological examination, he was confused; he could speak but could not give accurate answers to questions. There was no motor impairment or meningeal sign. Routine laboratory investigation findings were normal including venous blood gas, blood lactate, glucose, electrolytes, liver and kidney function tests, TSH, free T4, ammonia. The brain magnetic resonance imaging and cerebrospinal fluid examination showed no abnormality. Electroencephalography (EEG) showed high-voltage slow wave activity on the right hemisphere with epileptiform discharge. Spike-slow wave complex discharge was seen with 1-3 Hz frequency on the right temporoparietal region (Figure 1). The ultrasound examination on the second day was abnormal with fluid in the abdominal cavity. He had immediately undergone surgery. At exploration, the peritoneal cavity was filled with serohemoragic fluid. The appendix and cecum was normal. A torsed and gangrenous Meckel's diverticulum with extension of ischemia to adjacent small bowel was seen and resected. His consciousness had become normal by the third day. The EEG on the sixth day was normal. He was discharged on the seventh day after operation without any sequela.

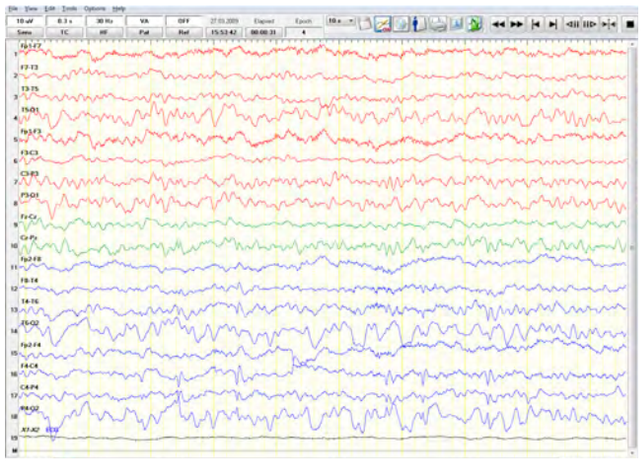

Figure 1. The EEG of the patient on the second day showing high-voltage slow wave activity on the right hemisphere with epileptiform discharge on the right temporoparietal region

Discussion

Sepsis-associated encephalopathy is a reversible dysfunction of the central nervous system that has a wide spectrum of clinical presentations from stupor and coma to severe agitation and irritability [2].

The pathogenesis of SEA is likely to be multifactorial [1]. Encephalopathy is not directly due to the toxin release from a microorganism. SEA is more likely caused by the mediators of inflammation (such as interferon alpha, interleukin 1 beta or tumor necrosis factor-alpha) which have a direct effect on the function of neurons and the blood-brain barrier. Also microcirculatory failure, activation of complement system, mitochondrial dysfunction alter brain function [2].

Evidence of extracranial infection with impaired mental state is necessary for the diagnosis of SEA. Sometimes finding the focus of extracranial infection may be difficult. So intracranial infections such as encephalitis, meningitis and brain abscess should be excluded by using lumbar puncture, computed tomography or magnetic resonance imaging [1]. In our patient, we also excluded intracranial infection. Our patient's brain magnetic resonance imaging and cerebrospinal fluid examination showed no abnormality. There is no diagnostic test with high specificity for SEA. Blood culture positivitiy is less than 50% of SEA patients [1]. Brain imaging methods such as computed tomography or magnetic resonance imaging are not definitive. Electroencephalography (EEG) is the most sensitive diagnostic tool for SEA and especially useful in the intensive care monitoring of patients with sepsis [7]. Also disturbances detected by EEG correlate with the severity of SEA [2,7]. Generally, the electroencephalogram findings of the patient with SEA demonstrate diffuse cerebral dysfunction [7-9]. Hosokawa et al reported that the incidence of EEG abnormalities during sepsis ranged from 12% to 100% for background abnormality and 6% to 12% for presence of triphasic waves. Also in patients with SEA, epileptiform discharges and electrographic seizures were also defined [10]. In our patients, EEG showed right hemisphere dysfunction with epileptiform discharge.

In conclusion, SEA is the one of major complication of sepsis and has a high mortality rate. Also neurodevelopmental and behavioral outcomes are poor. The children with SEA have delayed neurodevelopment, low verbal IQ, decline in school performance and low intelligence at short-term follow-up [11]. To overcome a poor prognosis in patients with SEA, the early recognition of the symptoms of SEA and also appropriate treatment of the underlying cause are essential. Electroencephalography is a diagnostic tool which helps us for determining the mental status of the patient.

Competing interests

The authors declare that they have no competing interests.

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Nonsustained Repetitive Upper Septal Idıopathic Fascicular Left Ventricular Tachycardia: Rare Type of VT

Tekrarlayan Süreksiz İdiyopatik Sol Ventriküler Üst Septal Fasiküler Taşikardi: Nadir Tip VT

Upper Septal Fascicular Left Ventricular Tachycardia

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Özet

Üst septal fasiküler ventriküler taşikardi, idiyopatik fasiküler ventriküler taşikardi subtipleri arasında oldukça nadir görülen bir taşikardidir. Üst septal fasiküler taşikardi anterograd yolunda posterior fasikülü, retrograd yolunda ise septal fasikülü kullanmaktadır. Dar QRS morfolojisi ve normal aks özelliğine sahip bu taşikardinin elektrokardiyografik bulguları supraventriküler taşikardi ile karıştırılabilmektedir. Biz burada His-Purkinje sisteminin proksimalinden kaynaklanan oldukça nadir bir fasiküler taşikardi alt tipini raporladık.

Anahtar Kelimeler

Üst Septal Fasiküler Taşikardi; Ablasyon; Purkinje Potansiyel

Abstract

Upper septal fascicular ventricular tachycardia is a very rare form of idiopathic fascicular ventricular tachycardia. Upper septal fascicular tachycardia uses the posterior fascicle as the anterograde limb and the septal fascicle as the retrograde limb. When evaluating the electrocardiography for this form of tachycardia, the presence of narrow QRS morphology and normal axis may be misinterpreted as supraventricular tachycardia. Here, we report a very rare subtype of fascicular tachycardia that originates more proximally in the His-Purkinje system at the base of the heart.

Keywords

Upper Septal Fascicular Tachycardia; Ablation; Purkinje Potential

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Introduction

Idiopathic fascicular ventricular tachycardia (VT) of the left ventricle is a rare type of VT. It occurs predominantly in young males (15-40 years old). Unlike idiopathic right ventricular outflow VT, which usually occurs as either nonsustained or sustained monomorphic VT, idiopathic fascicular left VT usually occurs as sustained monomorphic [1]. Verapamil-sensitive fascicular VT is the most common form of idiopathic left VT. The mechanism of this tachycardia is reentry within the left-sided specialized conduction system. The most common form of these tachycardias affects the posterior fascicle and is therefore known as posterior fascicular VT. Left posterior fascicular VT exhibits right bundle branch block (RBBB) morphology and superior axis. A less common form, left anterior fascicular VT shows RBBB morphology and right axis. A very rare form affecting the septal fascicle, known as upper septal fascicular VT, has narrow QRS morphology and normal or right axis deviation [2-4].

Case Report

A 46-year-old female patient was admitted to the cardiology outpatient clinic with symptoms of intermittent attacks of palpitations and discomfort. An initial electrocardiogram (ECG) revealed that she had sinus rhythm with repetitive nonsustained 185-190 bpm narrow QRS complex tachycardia without an axis deviation (Figure 1). The patient's complete blood count (CBC),

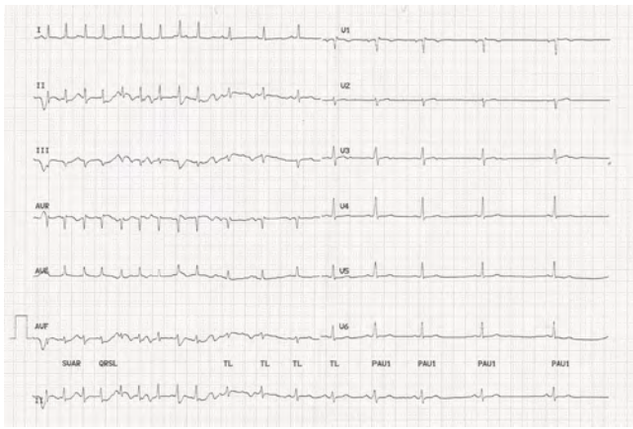

Figure 1. Twelve-lead electrocardiogram of nonsustained fascicular ventricular tachycardia with narrow QRS morphology and normal axis

renal function tests, thyroid function tests, and serum electrolyte levels were normal. Echocardiography was performed and left ventricular systolic and diastolic functions were normal. There was no evidence of structural heart disease. The patient used B-blocker medicine at times when she had palpitation attacks; she refused the chronic antiarrhythmic treatment. We offered a diagnostic electrophysiological study and planned an ablation procedure.

A quadripolar electrophysiology and radiofrequency ablation catheter (7F Marinr ablation catheter, Medtronic, USA) were used via the right femoral vein and artery. In the electrophysiological study, AH-HV intervals were normal. Simultaneously with the catheter manipulations, a sustained 187 bpm narrow QRS tachycardia with normal axis that was almost identical to the clinical tachycardia was induced (Figure 2). Atrioventricular dissociation (AV) was observed during tachycardia (Figure 3). Clinical tachycardia was easily induced with programmed

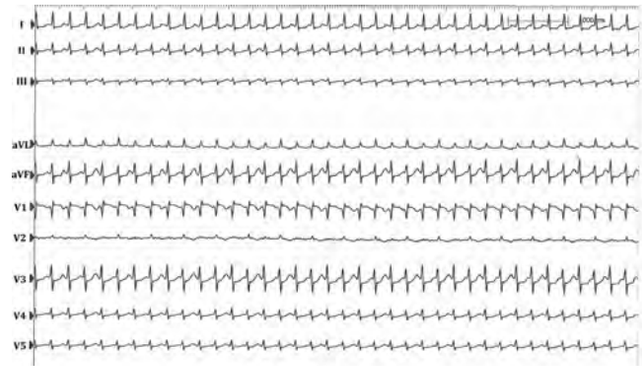

Figure 2. Twelve-lead electrocardiogram of sustained upper septal idiopathic fascicular left ventricular tachycardia

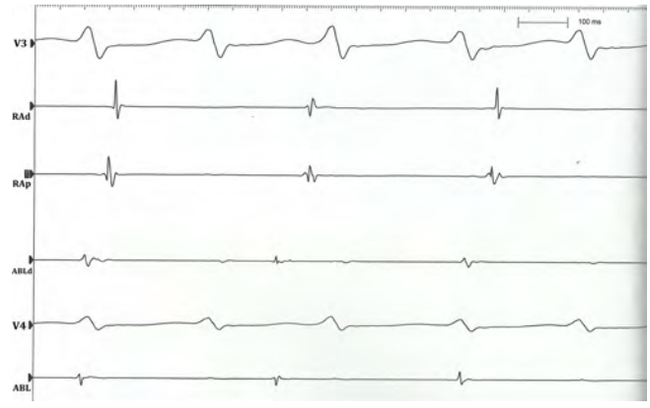

Figure 3. Intracardiac recordings during upper septal fascicular tachycardia with atrioventricular dissociation

electrical stimulations (PES) from right ventricular apex (RVA). Tachycardia was not induced with PES from high right atrium (HRA) under isoproterenol infusion. During VT, retrograde activation of the His bundle was recorded before the onset of the QRS complex with a His-ventricular interval that was shorter during VT than that during sinus rhythm (26 msn vs. 51 msn respectively). Endocardial mapping was made by retrograde transaortic approach within the left ventricle. Early Purkinje potentials before the onset of QRS were detected by an activation mapping technique during tachycardia. At the upper ventricular septum activation mapping during tachycardia, at the level od distal His bundle or proximal left bundle branch, early Purkinje potential was observed 23 msn before the onset of QRS (Figure 4). During tachycardia, five radiofrequency ablations were performed to the site of earliest Purkinje potential (40 W, temperature 42°C -52°C). Due to the proximity to the His bundle, RF ablations were performed carefully and for a short time. After the procedure, clinical tachycardia was not inducable with PES from RVA. Following the procedure, a twelve-lead ECG revealed no AV complete block and left bundle branch block (LBBB) (Figure 5). Following the RFA procedure, the tachycardia could not be reinduced, stable sinus rhythm without bursts of VT was maintained, and baseline QRS morphology remained unchanged. After a follow up of 3 months without antiarrhythmic drugs, the patient was asymptomatic without new episodes of tachycardia.

Discussion

Ventricular tachycardias spreading from the anterior and posterior divisions of the left bundle branch are generally called fascicular tachycardia. Fascicular tachycardia has been classified

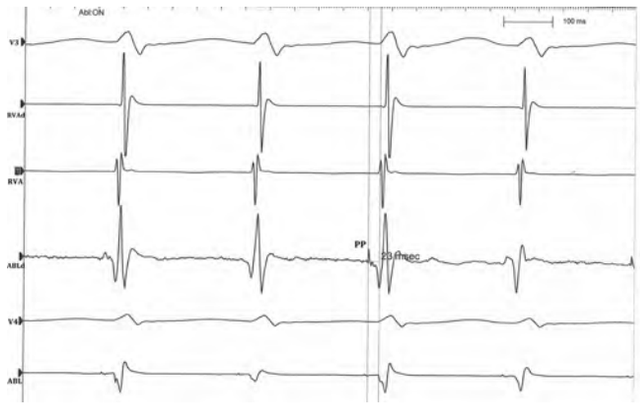

Figure 4. Purkinje potential (PP) preceding the onset of QRS during fascicular tachycardia at the successful ablation site at the left upper septum

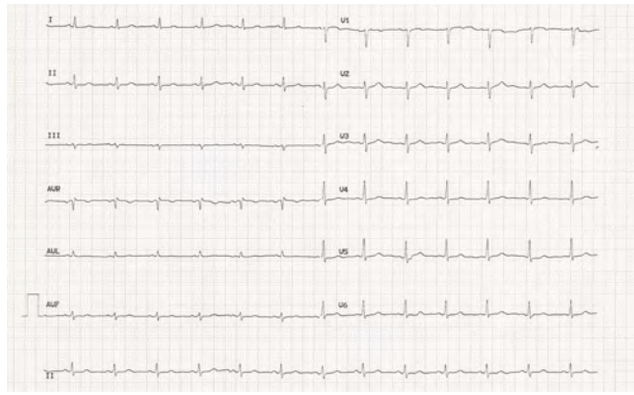

Figure 5. Twelve-lead electrocardiogram after tachycardia ablation

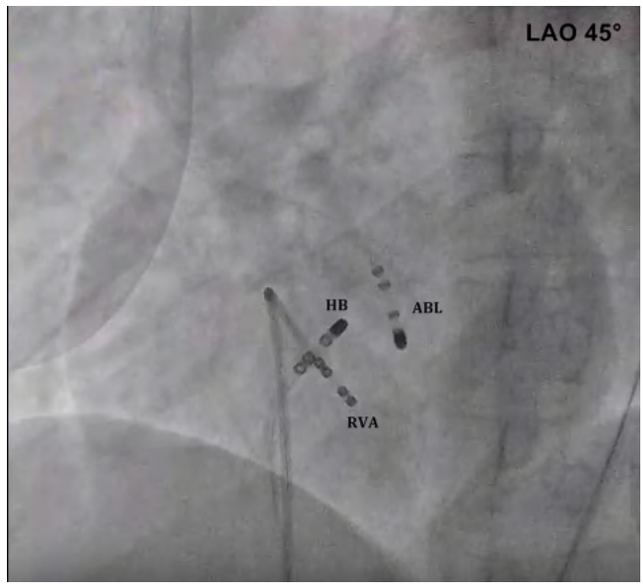

Figure 6. Fluoroscopy of catheter position at successful ablation site

into three subtypes. First, left posterior fascicular VT with right bundle branch block (RBBB) pattern and superior axis deviation. Second, left anterior fascicular VT with RBBB pattern and right axis deviation. Third, upper septal fascicular VT with a narrow QRS and normal axis configuration. Upper septal tachycardia usually exhibits incomplete RBBB morphology [4]. Rare cases exhibiting LBBB (precordial R wave transition between V3 and V4) and a normal frontal plane axis have been reported [5]. The most common form of fascicular tachycardia is the posterior fascicular type, which accounts for nearly 90% of the cases. Left anterior fascicular VT is uncommon (approximately 10%), and left upper septal fascicular VT is very rare (less than 1%). Overwhelming evidence suggests that idiopathic fascicular left ventricular tachycardia is caused by a re-entrant circuit incorporating the Purkinje system with an excitable gap and slow conduction area [4]. Upper septal idiopathic fascicular tachycardia uses portions of the posterior fascicular normal Purkinje fibers as the anterograde limb (which can be considered an orthodromic form of posterior fascicular VT) and the septal fascicular abnormal Purkinje fibers as the retrograde limb. There is simultaneous passive activation of the right bundle branch and anterior fascicle; this accounts for the relatively narrow QRS, which can be very similar to baseline QRS [4]. In our patient, the QRS morphology of the tachycardia was similar to the baseline QRS morphology of the twelve-lead ECG at admission. Upper septal fascicular tachycardia is often associated with a history of ablation of a typical posterior fascicular VT, although our patient had not undergone this procedure. The fascicular tachycardia circuit can be constructed from the observations during VT. During fascicular tachycardia two distinct potentials can be observed before the ventricular electrogram, namely the Purkinje potential (PP) and pre-Purkinje potential (pre-PP), also designated P2 and P1 respectively. In upper septal fascicular tachycardia, Purkinje potentials were activated in a reverse direction to that of left posterior fascicular tachycardia; namely, pre-Purkinje potential (P1) was activated retrogradely but the Purkinje potential (P2) was activated anterogradely. Purkinje potential (PP or P2) , first described by Nakagawa et al. [6], represents the activation of the left posterior fascicle or the Purkinje fibers near the left posterior fascicle; this potential precedes the onset of QRS during tachycardia. The pre-Purkinje potential (pre-PP or P1) was first described by Tsuchiya et al. [7]. It represents excitation at the entrance to the specialized zone in the ventricular septum which has decremental properties and is sensitive to verapamil. Pre-PP potential precedes PP potential during fascicular tachycardia. Nogami et al. [8] observed that the interval between PP at the site of successful ablation and the onset of the QRS complex during VT was 18±6 msn (6±3% VT cycle length). In our case, at the upper ventricular septum activation mapping during tachycardia, early Purkinje potential was observed 23 msn before the onset of QRS. At this point in the procedure, it was possible to entrain the tachycardia. A difference was obtained between the post-pacing interval (PPI) and tachycardia cycle length (0 msn), with an interval between the peak pacing level and the QRS onset equal to the interval between the fascicular potential and QRS onset. These findings indicate that there was a reentry mechanism and the ablation catheter was positioned at the site of the tachycardia circuit. Finally, in such cases, ventricular tachycardias reflecting RBBB morphology, such as mitral anular VT, intramyocardial reentry VT, and interfascicular VT, should be considered in the differential diagnosis. The main cause for arrhythmia in this case was the reentry mechanism associated with slow entrance of calcium into Purkinje fibers.

Competing interests

The authors declare that they have no competing interests.

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Evaluation of Heterozygous Deletion of TP53 Gene in Pleural Fluid Samples: A Case Series of 11 Patients

Plevral Sıvı Örneklerinde TP53 Geni Heterozigot Delesyonunun Değerlendirilmesi: 11 Hastalık Bir Seri

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Özet

Biz, malign hastalığı olmayan 2 hasta (pnömoni) ve malign hastalığı olan 9 hastada [Küçük hücreli akciğer karsinomu (n=3), küçük hücreli dışı akciğer karsinomu (n=4), non-hodgkin lenfoma (n=1) ve mide kanseri (n=1)] Tümör Protein 53 (TP53) genindeki heterozigot delesyonu tanımladık. Kromozomal aberant durum, sentromer ve 17p13.1 lokusuna özellikli problar kullanılarak floresan in situ hibridizasyon yoluyla analiz edildi. Dokuz kanser hastasının 3'ünde, histolojik değerlendirme ve/veya kapalı plevral biyopsi ile malign plevral efüzyon tespit etmedik. TP53 geni heterozigot delesyonu malign hastalığı olanlarda bening plevral efüzyonu olanlara kıyasla belirgin olarak yüksek olarak bulundu. Sonuç olarak, TP53 heterozigot delesyonu malignansi için bir belirteç olabileceğini öne sürmekteyiz, bununla birlikte bu öneriyi desteklemek için büyük hasta kohortları ile ileri çalışmalar gerekmektedir.

Anahtar Kelimeler

Plevral Efüzyon; Malignite; TP53 Gen

Abstract

We described heterozygous deletion of tumor protein 53 (TP53) gene in 11 patients including 2 patients with non-malignant diseases (pneumonia) and 9 patients with malignant diseases [including small cell lung cancer (n = 3), non-small cell lung carcinoma (n = 4), non-Hodgkin's lymphoma (n=1), and gastric carcinoma (n=1)]. Chromosomal aberrant status was analyzed by fluorescence in situ hybridization with centromere specific and 17p13.1 locus specific probes. In 3 of 9 cancer patients we did not find malignant pleural effusion with histological examination and/or closed pleural biopsy. Heterozygous deletion of TP53 gene was found to be significantly higher in patients with malignant disease when compared to the patients with benign pleural fluid. As a result, we suggest that heterozygous deletion of TP53 may have indicator value for malignancy; however further studies are warranted to confirm this suggestion in large patient cohorts.

Keywords

Pleural Effusion; Malignancy; TP53 Gene

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Introduction

Malignant pleural effusion (MPE) is a common complication in patients with advanced cancers, occurring in 15% of cancer-related deaths. MPE is thought to be caused by the hyper permeability of microvascular tissue or invasion of cancer cells into lymphatic vessels. Most pleural effusions (PEs) occur with concomitant tumor. However, in a few patients, multiple cytopathologic examinations of pleural fluid are negative for tumor [1,2]. Differential diagnoses of malign and benign pleural effusions remain a challenge. The accuracy of cytological examination of the diagnosis PEs is about 60% and pleural biopsy contributes 7–13% [3]. Video-assisted thoracic surgery is the gold standard procedure in the diagnosis of PEs; however surgical procedures may have several complications. Therefore, avoiding invasive procedures with several serious complications lead clinicians to try to find non-invasive tests such as pleural fluid biomarkers, particularly through the genetic analysis of PEs.

In humans, TP53 gene is located on the short arm of chromosome 17 (17p13.1) [4]. TP53 is the most altered gene in cancer. TP53 mutation was found to be in more than 50% of human cancers. Mutations disabling the TP53 tumor suppressor gene represent the most frequent events in human cancer and typically occur through a double-hit mechanism involving a missense mutation in one allele and a "loss of heterozygosity" deletion encompassing the other [5]. Deletion of TP53 gene has not been reported to date in the diagnosis of malignant pleural effusion. In the present study, we reported numerical chromosomal status in effusion cells derived from 11 patients with malignant and non-malignant diseases by using fluorescence in situ hybridization (FISH) with centromere specific and 17p13.1 locus specific probes for chromosomes 17.

Material and Method

The present study was carried out at Süleyman Demirel University Medical Faculty. All patients were male. Eleven pleural effusion specimens were derived from 11 patients including 2 patients with non-malignant and 9 patients with malignant diseases. In 3 of 9 patients with malignancy, the pleural effusion samples showed non-malignant feature. Closed pleural biopsy or medical thoracoscopy performed in these 3 patients indicated there was no malignancy in pleural fluid. In all these malignant cases, metastatic diseases were defined by computed tomography (CT) or magnetic resonance imaging (MRI). Cytological evaluation was also performed in each sample.

Fluorescence in situ hybridization

Fluorescence in situ hybridization targeting TP53 locus was performed for pleural fluid samples. A direct fluorochrome-labeled, dual-color DNA probe cocktail (Cytocell, UK) for P53 -17p13.1 and reference loci (D17Z1) was hybridized to slides of pleural fluid samples. Hybridizations and washings were carried out according to the stringency conditions and the procedures recommended by the manufacturers. Dual-color fluorescent signals were detected and analyzed under epifluorescence microscopy equipped with specific filter sets. At least 200 interphase nuclei were analyzed and scored by independent investigators.

Case Series

The key features of these cases are summarized in Table 1. The mean age was found as 75.3 years. Of the 9 malignant patients, 3 patients have small cell cancer, 4 patients have non-small cell cancer, 1 patient has non-Hodgkin's lymphoma, and 1 patient has gastric cancer. The diagnosis of 2 patients with benign disease was pneumonia. There were lower levels of heterozygous deletion in TP53 gene in patients with benign disease (cases 10-11) and patients with malign disease non-malignant pleural fluid (cases 1-3) when compared to patients with malignant pleural fluid (cases 4-9). TP53 gene deletion ratio in patients with benign pleural effusions was detected at less than 10% while the ratio in patients with malign effusion was found over 10% (table 1, figure 1).

Table 1. The Heterozygous Deletion of P53 and Patients Characteristics

Case	Age	Origin/Histology/Stage	Pleura Fluid	Heterozygous Deletion of p53 Gene (%)
1	59	Small cell cancer, Limited stage	Non-malignant	3.6
2	60	Small cell cancer, Advanced stage	Non-malignant	1.2
3	61	Non-small cell cancer, adenocarcinoma, stage IIIB	Non-malignant	5.9
4	73	Gastric adenocarcinoma, stage IV	Malignant	9.2
5	66	Small cell cancer, Advanced stage	Malignant	17.2
6	58	Non-small cell cancer, adenocarcinoma, stage IV	Malignant	36.2
7	67	Non-small cell cancer, NOS, stage IV	Malignant	25.2
8	79	Non-Hodking's Lymphoma	Malignant	15.3
9	86	Non-small cell cancer, adenocarcinoma, stage IV	Malignant	2.1
10	56	Pneumonia	Non-malignant	0.65
11	88	Pneumonia	Non-malignant	1.35

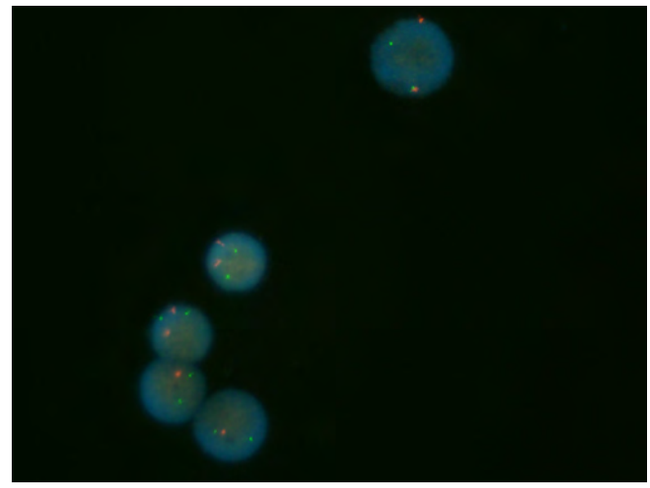

Figure 1. Interphase fluorescence in situ hybridization (FISH) results. FISH using P53 (TP53) gene deletion probe, reveals Green(G); chromosome D17Z1 locus, Red(R); chromosome 17p13.1 (P53 gene). Blue; DAPI. Normal nuclei: 2G/2R, Nuclei with P53 deletion: 2G/1Rv

Discussion

Cancer is one of the main causes of exudative PEs. Many of the biochemical markers in pleural fluid are currently being investigated and/or utilized in the clinic. However, the differential diagnosis is still difficult because the mechanism of PE formation

is multifactorial and not completely understood. Furthermore, detecting malignant and non-malignant effusion is still a challenging issue; biological behavior and differentiation of each malignant cell are different. For this purpose, genetic studies of pleural effusion have been reported [6]. But the detection of TP53 gene heterozygous deletion in pleural fluid has not been reported to date. Therefore in the present paper, we hypothesized that heterozygous deletion of TP53 gene could be a beneficial candidate in the differential diagnosis of MPEs.

We found higher levels of heterozygous deletion of TP53 in PEs of malignant origin than in PEs of benign origin. However, malignant patients with benign pleural effusion had TP53 gene deletion similar to PEs of benign origin (table 1). Cora et al, found that there is an association between chromosomes aneuploidies and pleural effusion cell status [6]. The FISH method was used in this study. In another genetic study of pleural fluid, p16 gene homozygous deletion in pleural effusion was found to be strongly related to malignant status and higher metastatic potential [7]. The same method was used for genetic testing in this study.

Fluorescence in situ hybridization is a cytogenetic technique that uses fluorescent probes that bind to only those parts of the chromosome with a high degree of sequence complementarity. Its role seems to increase in detecting the characterization of chromosomal rearrangements and marker chromosomes, the detection of micro deletions, and the prenatal diagnosis of common aneuploidies [8-10]. By using an interphase-FISH technique, specific genetic aberrations have been reported in effusion samples of patients with cancer [11,12]. The FISH technique, as seen in our series, may be a safe method in the differential diagnosis of malignant and benign pleural effusion.

P53 is the most altered gene in cancer. More than 50% of human cancers are afflicted with a TP53 mutation. The previous studies suggested that the use of p53-antibodies has potential diagnostic value for several cancers [13,14]. But TP53 mutation has not been evaluated in body fluid samples for cancer diagnosis. For this reason our results may be important. However, the number of cases is quite small and it is not a homogeneous group. Despite these limitations, we believe that future genetic studies may be planned in the light of the present report.

Competing interests

The authors declare that they have no competing interests.

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Pleomorphic Adenoma of the Nasolabial Region

Nazolabial Bölgenin Pleomorfik Adenomu

Nasolabial Pleomorphic Adenoma

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This study was presented as an poster presentation (number: PS-1031) at the 37th congress of the Turkish National Otorhinolaryngology and Head & Neck Surgery, September 28th-November 01, 2015, Kaya Plazzo Hotel, Antalya, Turkey.

Özet

Pleomorfik adenom genellikle büyük tükürük bezlerinde, en sık parotis bezinde oluşur. Nazal kavite, nazofarenks, trakea ve özefagus gibi alışılmadık pleomorfik adenom vakaları bildirilmiş olmasına rağmen, bizim bildiğimiz kadarıyla bu mevcut vaka nazolabial bölgede ilk rapor edilendir. Bu çalışmada, bu nadir lezyonun klinik görünümü, cerrahi bulguları, histopatolojik özellikleri ve tedavi zorlukları tartışılmıştır.

Anahtar Kelimeler

Pleomorfik Adenom; Tükürük Bezi Tümörleri; Baş Boyun Tümörleri

Abstract

Pleomorphic adenoma often occurs in the major salivary glands, most commonly on the parotid gland. Although unusual cases of pleomorphic adenoma of the nasal cavity, nasopharynx, trachea, and oesophagus have been reported, to the best of our knowledge the present case is the first reported case in the nasolabial region. In this article, clinical presentation, surgical findings, histopathological features, and therapeutic challenges of this rare lesion are discussed.

Keywords

Pleomorphic Adenoma; Salivary Gland Tumors; Head And Neck Neoplasms

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Introduction

Pleomorphic adenoma (PA), the most common type of benign salivary gland tumor, histopathologically consists of epithelial and stromal components. PAs usually appear as solitary, painless, asymptomatic, and slow-growing lesions. Though it is classified as a benign tumor, PA may undergo malignant transformation into carcinoma ex-pleomorphic adenoma, a risk that is believed to increase with time [1]. Also it can recur after resection; distant metastases have been reported after long time intervals [2, 3].

PA often occurs in the major salivary glands, most commonly on the parotid gland [4]. The palate is the most affected site among the extra-major salivary glands [3, 5]. The nasolabial region (NLR) is an unexpected involvement site for these tumors. Our study represents a case of a PA of the NLR excised by an intraoral (sublabial) approach. In this article, clinical presentation, surgical findings, histopathological features, and treatment of this rare lesion are discussed.

Case Report

A 21-year-old female presented with a complaint of a painless lump, having gradually increased in size, in the right NLR. She had first noted the lesion about nine months previously. The patient reported no trauma. On palpation the lesion was elastic, well-circumscribed, movable, painless, and 1.5 to 2 cm in diameter. Bimanual palpation of the upper lip was normal. The overlying skin and labial mucosa was not fixed. She was found to have a mild, barely noticeable asymmetry of the face. No other abnormality was noted on otolaryngologic examination. She had no significant previous medical history. The mass was totally excised with an intraoral (sublabial) approach. An incision was made in the oral cavity mucosa along the gingivolabial sulcus under local anesthesia. Dissection exposed a smooth, well-circumscribed, encapsulated solid mass that was approximately 2 cm in diameter (Figure 1). No adhesion to the underlying bone was encountered. The lesion was excised without difficulty from the surrounding tissue. The specimen obtained was a gray-white lesion, fully encapsulated with normal margin (Figure 2). The wound was closed by primary intention. The specimen was fixed in 10% buffered formalin and submitted for histopathologic diagnosis. The postoperative course was uneventful. No additional treatment was required. Subsequent follow-up after three years showed no signs of recurrence. Histologically, the material showed a well encapsulated epitelial salivary gland tumor. The lesion had a thin fibrous capsule and consisted of epithelial and fibromyxoid stromal components. A diagnosis of PA was made based on the characteristic histological pattern (Figure 3).

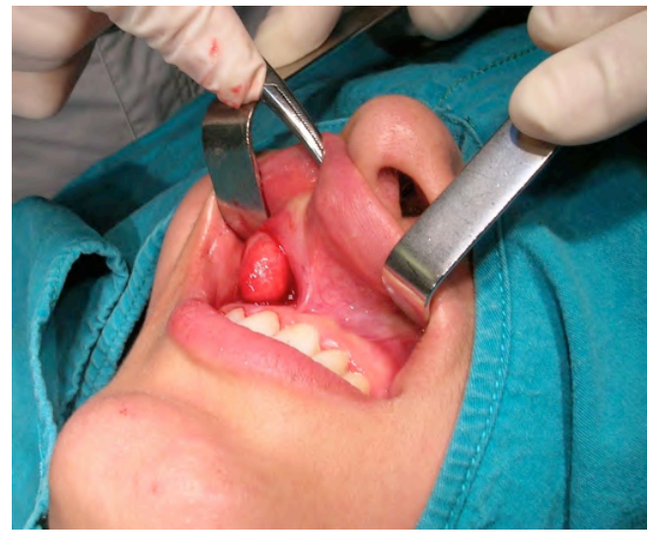

Figure 1. The tumor, after fully released from the surrounding tissue and pulled downwards.

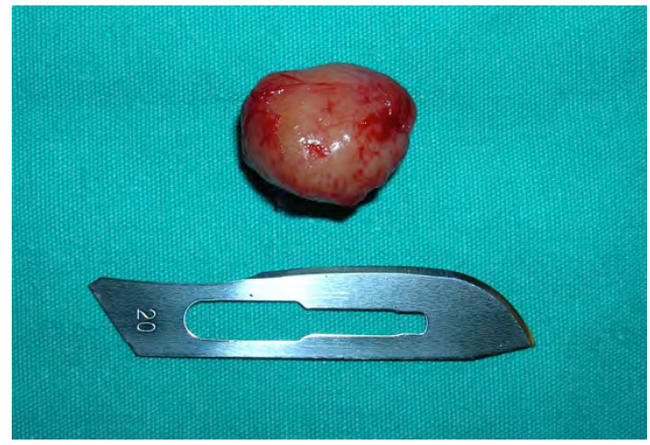

Figure 2. Specimen removed.

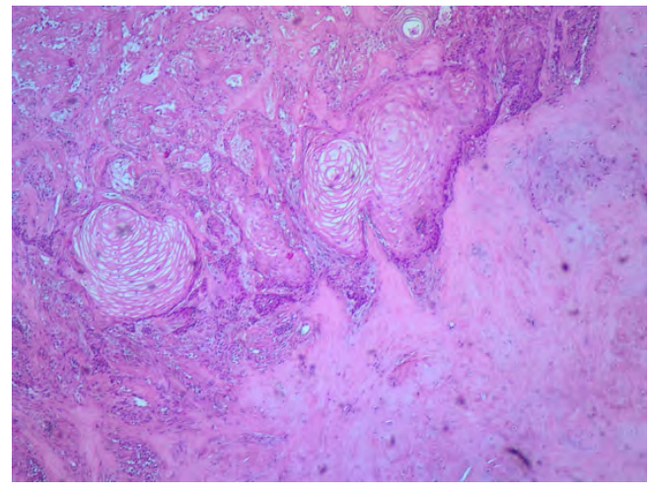

Figure 3. Histological pattern (H-E x 100).

Discussion

Our study presents a PA of the NLR causing a painless mass as the primary symptom. Although unusual cases of PA of the nasopharynx, orbital area, trachea, nasal septum, and external auditory canal have been reported, to the best of our knowledge the present case is the first to be reported in the NLR [5]. Based on the clinical appearance, the preliminary diagnosis in our case was a nasolabial cyst, the most likely tumor found in this region. Nasolabial cysts are frequently-asymptomatic developmental masses occurring inferior to the nasal alar region. A differential diagnosis of foreign body granuloma, lipoma, neurofibroma, myoepithelioma, or other benign mesenchymal tumors was also considered. A salivary gland tumor was not considered because of its deep location. A fine needle aspiration biopsy may be performed, but, due to the probable benign nature of the lesion, an excisional biopsy was planned. In our case, PA may have arisen from minor salivary glands of the nasal alar region and expanded to the NLR, possibly explaining the unusual location of the PA.

Tumors arising from the minor salivary glands are uncommon clinical entities. Limited information is available in the literature on the management of PAs located in the extra-major salivary glands. In general, it is suggested that the lesion be removed together with the surrounding normal tissue for PA treatment [3-5]. However, wide excision of extra-major salivary gland PA may not always be possible due to aesthetic concerns, therefore resulting in a close surgical margin. In their study, Kuo et al. retrospectively investigated 37 patients who had undergone primary surgery for extra-major salivary gland pleomorphic adenoma of the head and neck region and concluded that although adequate surgical margins were not always achieved during operation, the rate of recurrence was relatively low in the 4.5- year average follow-up period [5]. In the present case, total excision by a narrow margin via a sublabial approach was performed. The lesion was easily released from the surrounding tissue with clinically normal margins and the mass appeared to be fully encapsulated. Follow-up three years after surgery showed smooth healing of the wound with no evidence of recurrence. But considering that the capsule may have been infiltrated by extension of the tumor, the patient will be followed up for at least 10 years due to the risk of recurrence.

In conclusion, PA should be considered in the differential diagnosis of tumors located in the NLR. Wide surgical excision may not be always possible to achieve with adequate surgical margins because of aesthetic concerns in this region. Special surgical attention must be paid not to rupture the capsule during the operation. Long-term follow-up of patients is necessary due to the risk of recurrence.

Competing interests

The authors declare that they have no competing interests.

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Case Report
JCAM

Cesarean Delivery with Bilateral Ovarian Transposition for Locally Advanced Cervical Cancer

Lokal İleri Evre Serviks Kanserinde Sezeryan Doğumda Bilateral Ovaryan Transpozisyon

Locally Advenced Cervical Cancer in Pregnancy

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Özet

Biz vakamızda özellikle 3. trimestırda lokal ileri evre serviks kanserinin, sezeryan ile doğum esnasında ovaryan transpozisyon uygulama yöntemi hakkında tartışmayı amaçladık. Hastamız 26 yaşında gebeliğinin 29 haftasında olup 9 gündür amnios sızıntısı devam etmektedir. Steril spekulum incelemesi sırasında geniş servikal lezyon saptandı. Servikal Biyopsi sonucu invaziv servikal kanser olarak rapor edildi. Manyetik rezonans görüntülemede şüpheli sağ parametriyal katılımı ile nodal tutulumu olmayan (MRG) 55X 63X 68 mm servikal lezyon, tutulum gösterilmiştir. Non-reaktif fetal durum ve şüpheli koryoamnionit ön tanısı ile elektif sezeryan planlandı. Otuzuncu gebelik haftasında elektif sezeryan ve bilateral ovaryan transpozisyon uygulandı. Pelvik radyasyon, eksternal brakiterapi ileri evre serviks kanseri için standart tedavi olarak uygulandı. Genç kadınlarda gebeliğin 3. trimestırında fetal matürite sağlandığında sezeryan uygulamasına ilave ovaryan transpozisyon uygulanması önerilmelidir.

Anahtar Kelimeler

Servikal Kanser; Ovaryan Transpozisyon; Gebelik

Abstract

We aimed to discuss about management of pregnancy with locally advanced cervical cancer, especially at the third trimester with ovarian transposition concomittant with cesarean delivery.A 26 year old patient who was at the 29th week of gestation had amniotic leakage for 9 days. During the sterile speculum examination, a large cervical lesion was detected.The cervical biopsy revealed invasive squamous cervical cancer. An magnetic resonance imaging(MRI) showed 55X63X68mm cervical lesion with suspicious right parametrial involvement, and no nodal involvement.An elective cesarean delivery decision was taken due to non-reassuring fetal status and suspicious chorioamnionitis.At 30th week of gestation, an elective cesarean delivery with bilateral ovarian transposition was performed.Pelvic radiation, including external beam and brachytherapy, has been the standard treatment of advanced cervical cancer.During the third trimester, fetal maturity is awaited and a caesarean section followed by standard treatment is proposed and the ovaries can be preserved with ovarian transposition in young women.

Keywords

Cervical Cancer; Ovarian Transposition; Pregnancy

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Introduction

Cervical cancer is relatively uncommon during pregnancy, however, it is the most commonly diagnosed gynaecological malignancy during pregnancy: incidence rates vary from 0.1 to 12 per 100,000 pregnancies[1,2]. The managament is difficult and some individualized factors such as diagnosed time of gestational age, patient desire, tumor size, diseases stage, histology, maternal and fetal risks can effect management. We present a case who had pregnancy with cervical cancer that was diagnosed at the third trimester. We aimed to discuss about management of pregnancy with locally advanced cervical cancer, especially at the third trimester with ovarian transposition concomittant with cesarean delivery

Case Report

A 26 year old patient who was at the 29 th week of gestation had amniotic leakage for 9 days. During the sterile speculum examination, a large cervical lesion was detected. The cervical biopsy revealed invasive squamous cervical cancer. She was referred to our clinic for follow-up and treatment. Speculum and pelvic examination revealed a 5 cm cervical mass on the anterior lip of the cervix, upper vaginal involvement and suspicious right parametrial involvement. The rupture of membranes (PPROM; preterm premature rupture of membrane) was confirmed. An magnetic resonance imaging (MRI) showed 55X 63X 68 mm cervical lesion with suspicious right parametrial involvement, and no nodal involvement (Figure1-2).

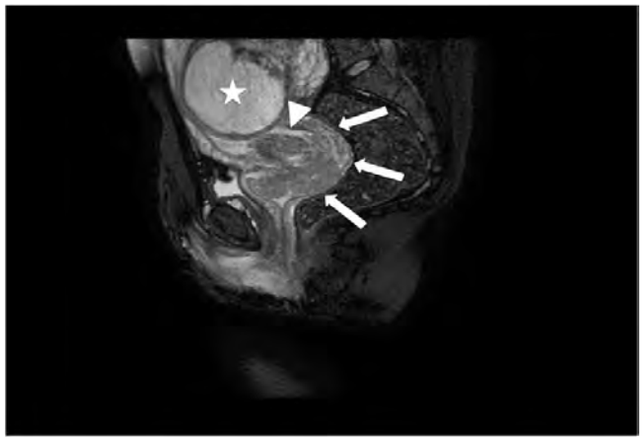

Figure 1. Sagittal T2 weighted MR image shows cervical carcinoma invading the cervical stroma and posterior fornix of the vagina (arrows). Upper part of the cervical canal (arrow head) can be normally seen. Fetal head (asterix) is located beside the cervix

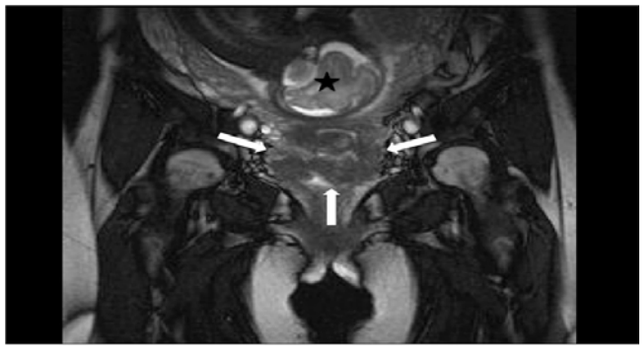

Figure 2. Coronal T2 weighted MR image shows cervical carcinoma invading the cervical stroma and minimally parametrium (arrows). Fetal head (asterix) is located above the cervix.

The fetal biometry and well being was evalauted. Delaying treatment to gain time for fetal maturity was choosen. A single course antenatal corticosteriod was given. The fetus and maternal status were evaluated regularly. The white blood and erythrocyte sedimentation rate (ESR) were 9000/ mm3, and 96 mm/h, respectively. An elective Cesarean delivery decision was taken due to non-reassuring fetal status and suspicious chorioamnionitis. At 30 th week of gestation, an elective cesarean delivery with bilateral ovarian transposition was performed. The patient was referred to radiotherapy after postpartum period. The baby had no abnormalities with follow-up ranging from birth to 10 months. The patient was still undergoing treatment when her case was reported.

Discusson

Cervical cancer is the most commonly diagnosed gynaecological malignancy during pregnancy. 70% of cervical cancers during pregnancy are diagnosed at stage I [3]. The most common symptom is vaginal bleeding, as well as vaginal discharge. Symptoms can be mistaken for complications of pregnancy, and diagnosis delay occurs if the level of suspicion is low. The diagnostic approach to cervical cancer during pregnancy is similar to that of nonpregnant women. Cervical cancer is staged clinically by clinical examination, in pregnant women, especially after the second trimester and beyond, clinical examination can be difficult[1,4]. Also, during pregnancy physiological cervical changes at cervical tissue such as oedem, increased vascularity, and hypertrophy may change tumor size, as well as stage.

For diagnosing and staging MRI can be used. MRI does not subject the fetus to ionizing radiation and may play an important role in the initial evaluation of the pregnant patient with cervical cancer. MRI can help determine tumour size in three dimensions, stromal invasion, vaginal and parametrial invasion, and also lymph node infiltration[5].

Multidisciplinary approach is recommended for treating a pregnant patient with cervical cancer. If pregnancy preservation is not aimed, management is similar to nonpregnant women. If pregnancy preservation is desired, the tretament choices can depend on before diagnosed 20- 22 weeks or after. For pregnant women with early stage disease (FIGO: IA2, IB1, 2A) diagnosed after 20 weeks of gestation, treatment may be delayed until the fetus has gain maturity[1,4]. An individualized treatment plan should be determined, regarding fetal maturity, however, delaying treatment beyond 32-34 weeks is not recommended. A Caesarean section may be chosen instead of vaginal delivery due to potential for haemorrhage, and the possibility of tumour implantation at episiotomy sites[6,7]. During pregnancy, surgery can be performed safely by skilled surgeons and anaesthetists. We started the operation with pfannenstiel incision and after cesarian section, peduncle of over was freed and was sutured outside the pelvis (Figure3).

When the pregnancy is terminated, the patient can be treated according to the stage of the disease. Pelvic radiation, including external beam and brachytherapy, has been the standard treatment of advanced cervical cancer. In younger women who referred to irradiation or radical hysterectomy, the ovaries can be preserved both hormonal and surrogate-assisted reproductive function[8]. Ovarian transposition is proposed as a way to

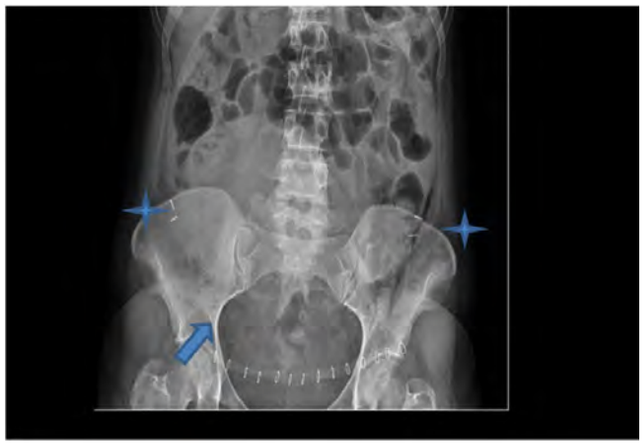

Figure. 3. The level of bilateral ovarian tansposition by marked metal clips at inferior ovarian edge.

preserve ovarian function in patients receiving pelvic radiation therapy.

The gestational week is the most important factor to decide for management of cervical cancer during pregnancy. During the third trimester, fetal maturity is awaited and a caesarean section followed by standard treatment is proposed. Multidisciplinary approach is mandatory regarding obstetrician, gynecologic oncology expert, radiation oncologist, neonatal specialist, oncologist, as well as with the patient and her relatives. There are no specific guidelines for the treatment of locally advanced cervical cancer during third trimester pregnancy. In young patients ovarian transposition during cesarean section is proposed as a way to preserve ovarian function in patients receiving pelvic radiation therapy.

Competing interests

The authors declare that they have no competing interests.

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Giant Fibrolipoma Extending Throughout a Whole Extremity: A Rare Child Case Report

Ekstremite Boyunca Uzanım Gösteren Dev Fibrolipom: Nadir Bir Çocuk Olgu Sunumu

Extremity Giant Fibrolipoma in Child

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Özet

Doğumsal olarak asimetrik büyüklükte alt ekstremiteleri olan 4 yaşında bir erkek hastayı sunmayı amaçladık. Olgunun doğum sonrasından itibaren yapılan radyolojik değerlendirmesinde, sol alt ekstremitesinde ekstremitenin yaş arttıkça olan büyümesine eşlik eden ve büyük oranda kalçadan itibaren ekstremite boyunca ayak düzeyine uzanım gösteren ve radyolojik modalitelerde geniş yağlı dokuyu işaret eden lezyon identifiye edildi. Ayak düzeyine yapılan cerrahi girişim ile eksize edilen dokunun histopatolojik değerlendirmesinde fibrolipom tanısı kondu. Bu, pediatrik hasta grubunda tüm ekstremiteyi kaplayan dev fibrolipom lezyonu olarak nadir bir durumdur.

Anahtar Kelimeler

Türkçe anahtar kelimeleri lütfen secretary@jcam.com.tr gönderiniz

Abstract

We present the case of a 4-year-old boy with congenitally asymmetrical lower extremities, his left being bigger than his right. The patient underwent imaging modalities after birth; a huge lipomatoid proliferation on his left thigh extended throughout the limb from the hip to the distal foot, showing isointensity with lipoid tissue. After surgical resection of his left foot, fibrolipoma was diagnosed. A giant fibrolipoma of the whole extremity occurring in a pediatric patient is a rarity.

Keywords

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Introduction

Lipomas are one of the most common benign mesenchymal soft tissue tumours of mature adipose tissue. Histologically, lipomas are classified into groups of simple lipomas, angiolipomas, angiomyolipomas, fibrolipomas, mixolipomas, lipomatosis, lipoblastoma, myelolipomas, spindle cell lipomas, hybernomas, and atypical lipomas, etc., according to the World Health Organization (WHO) classification [1]. Within these subtypes of lipomas, fibrolipoma is a rare subtype that is composed of adipose tissue separated by septations of connective tissue [2]. Fibrolipomas are commonly located subcutaneously; they can grow to huge dimensions or can become a nodular exophytic lesion. If there are cutaneous fibrolipomas caused by trauma, ischaemia and infarction lead to amorphous calcifications and fibrosis. They can also present with cartilaginous and/or osseous metaplasia [3].

In this report, we present a rare example of a giant fibrolipoma extending throughout a whole extremity in a child.

Case Report

Here we present the case of a 4-year-old boy with asymmetrical diameters of the lower extremities. His left thigh diameter had become thicker than the right one after birth. His family had also noticed that he could not wear the same shoe size, as his left foot was bigger. After clinical examination he underwent an imaging procedure. His anteroposterior radiograph (Figure 1) showed left thigh thickening, especially in the femoral side. Then on magnetic resonance (MR) images, a lesion overlying the left thigh from the hip to the foot distally was observed. On T1 and T2 weighted series (Figure 2,3), the lesion aspect showed hyperintensity; after fat saturation sequences (Figure 4), the lesion side showed signal loss, indicating lipoid tissue content. Histopathological results of a biopsy of the dorsal foot, where the lesion reached, indicated a fibrolipoma. Because of the lesion extending throughout the whole lower limb, it is named a giant fibrolipoma. Besides the radiological evaluation, laboratory tests and genetic analyses were applied but no correlation was found with any syndrome related to the lesion.

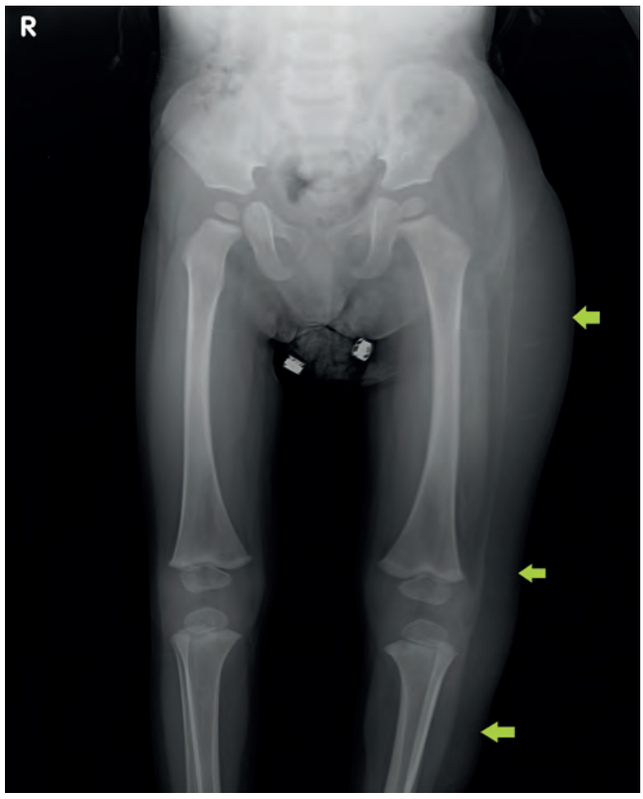

Figure 1. AP radiography image shows an increase in the left lower limb diameter, more in the upper side. Especially fatty tissue thickening in the lateral aspect.

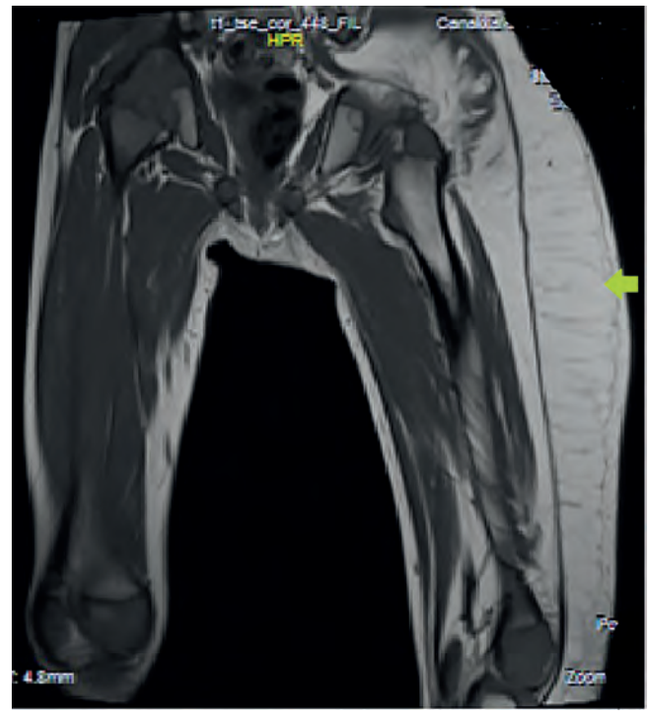

Figure 2. T1 weighted MR images; in coronal image lipoid tissue thickening seen in the lateral side of the femoral part of the thigh and in the sagittal image from foot and distal cruris.

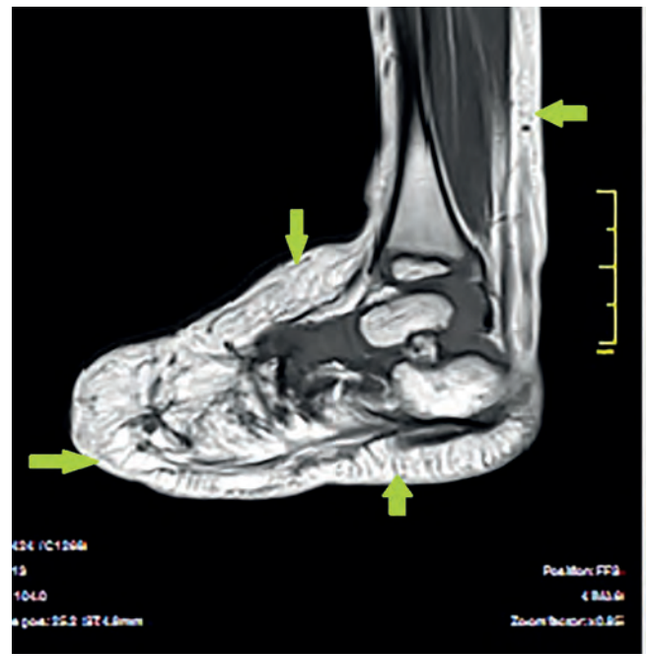

Figure 3. Sagittal T1 weighted MR images; thickening in the fatty tissue is shown, especially over the foot.

Discussion

Lipomas are tumours composed of mature adipocytes, mesenchymal primordial fat tissue cells, in the adipose tissue; they are one of the most common benign tumours. The etiology is not well known and they can be seen as sporadic cases or as related to inheritance [4]. Fibrolipomas, rare subtypes of lipomas,

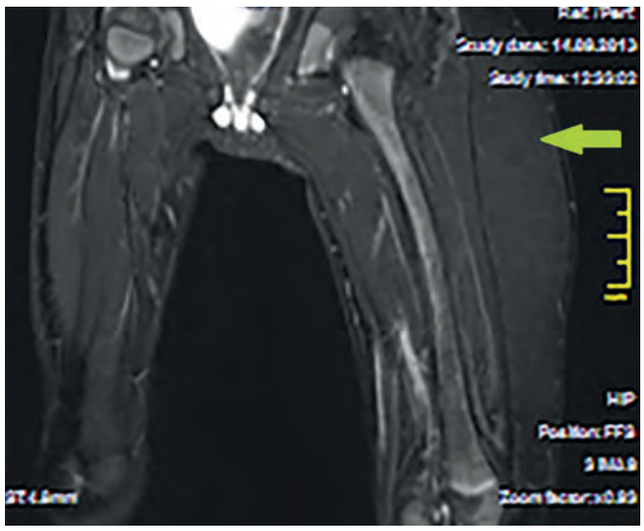

Figure 4. Coronal fat saturation proton density image shows hypointense fatty thickening in the lateral aspect of the femoral side in the left thigh.

may develop in the subcutaneous fat tissue, oral cavity, pharynx, larynx, esophagus, trachea, colon, parotid gland, and spermatic cord [5]. Although fibrolipomas may be seen anywhere in the body that contains fatty tissue, they commonly appear on the neck, trunk and upper limbs. Lower extremity localisation is not common [6]. Nerve fibrolipoma, spindle cell lipoma, subdermal fibrous hamartoma, mixolipomas, sclerosing liposarcoma, myolipoma, and nuchal fibroma should be considered in the differential diagnosis of fibrolipomas. Although they are known as benign neoplasms, the literature has reported some cases of liposarcoma, a malign transformation [7]. Giant lipomas of at least 10 cm in diameter or of a minimum weight of 1000 grams are most probably thought to be liposarcomas. The diagnosis of liposarcoma should be considered when the lesion is more than 10 cm in diameter and is growing quickly over a short period of time [8]. Histopathological criteria of invasion, mitosis, necrosis, and cellular atypia, together with the lipoblast, indicate malignancy.

The clinical presentation, treatment of choice, and the prognosis of fibrolipomas do not differ prominently from other types of lipomas. The management occasionally is surgical excision as a cure. Although they are known as benign tumours, the literature reports that there can be recurrences or malign transformations following the postoperative process [4].

In our case, postpartum macrodactyly was identified in the 2. Phalanx. During radiologic examination, fibro-adipose tissues on the plantar and dorsal areas of the foot were identified during the early postpartum period. After the child started to walk, he had difficulties with the movements of his left foot; he then underwent a surgical operation. Because of this, fibro-adipose tissue was excised from the plantar side localisation. Following this process, histopathological results of the excisional material indicated a diagnosis of fibrolipoma, a rare subtype of lipomas. Because the lesion extended throughout the whole extremity, from the hip to the distal foot, as the child grew, asymmetry in the lower extremities became more pronounced. This led clinicians in the orthopaedics department to make the decision to perform a surgical operation on his left extremity before he entered school.

As a result of this case, that of a rare giant fibrolipoma in a child, our clinical experience expanded. We want others to be aware that when lipomatoid tumours are suspected in a patient based on the clinical and radiological findings, histopathological examination should also be performed.

Competing interests

The authors declare that they have no competing interests.

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Early Plasmapheresis for Treatment of Acute Pancreatitis Related to Hypertriglyceridemia

Hipertrigliseridemiye Bağlı Akut Pankreatit Vakasında Erken Plazmaferez Uygulaması

Hypertriglyceridemic Acute Pancreatitis

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Özet

Akut pankreatit pankreasın ani inflamasyonu ile karakterizedir. Hipertrigliseridemi akut non-bilier pankreatitin önemli fakat genellikle atlanan, göreceli olarak nadir rastlanan bir nedenidir. Aferez çoğunlukla konzervatif tedaviye yanıt vermeyen hiperlipidemik olgularda uygulanır. Bu olgu sunumunda tip I hiperlipidemi ve tekrarlayan pankreatit atakları olan bir hastada erken plazmaferez uygulaması örneği verilmektedir.

Anahtar Kelimeler

Akut Nekrotizan Pankreatit; Hipertrigliseridemi; Plazmaferez

Abstract

Acute pancreatitis is characterized by a sudden inflammation of the pancreas. Hypertriglyceridemia is an important but usually unnoticed, relatively rare cause of acute non-biliary pancreatitis. Apheresis is mostly an option for hyperlipidemic cases failing to respond to conservative therapy. This study reports a case of acute necrotizing pancreatitis with a medical history of type I hyperlipidemia and recurrent attacks of pancreatitis for which early plasmapheresis was performed.

Keywords

Acute Necrotizing Pacreatitis; Hypertriglyceridemia; Plasmapheresis.

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Introduction

Acute pancreatitis (AP) is characterized by s sudden inflammation of the pancreas in which tissue architecture may be conserved (except interstitial edema and inflammatory cells in acute edematous pancreatitis) or destroyed by hemorrhagic and necrotic areas (in acute hemorrhagic/necrotizing pancreatitis). Inappropriate activation of pancreatic enzymes and then the autodigestion of pancreatic tisssue form the basis of the pathophysiology of AP. Focal or extensive fat necrosis can be revealed under a microscope in necrotizing form. Severity of the disease is based on clinical symptoms and laboratory and radiological findings. Mild disease cases usually recover uneventfully. However, local or systemic complications with organ dysfunction are inevitable if the disease is severe. Most cases are related to alcohol use or to biliary stones.

Severe hypertriglyceridemia is the most common cause of acute pancreatitis after biliary stones and alcohol use, according to the literature [1,2]. Hypertriglyceridemia is an important but unnoticed cause of acute non-biliary pancreatitis, responsible for approximately 3% of all acute pancreatitis cases [3]. On the other hand, acute pancreatitis occurs in 12-38% of hyperlipidemic patients [2,4]. Local excessive release of free fatty acids and lysolecithin from lipoprotein substrates exceeding the transport capacity of albumin, and therefore the development of injury in acinar cells and microvascular membranes, is the probable cause of acute pancreatitis in hyperlipidemic patients [5,6]. The main target of treatment is to lower serum triglyceride level and to repress the systemic inflammatory response. Patients with excessively elevated triglyceride levels need a fast and effective lowering of these levels in order to prevent a severe pancreatitis episode, so immediate apheretic treatment may be an option for a rapid lowering of these levels. Recently, the use of plasmapheresis for treating patients with severe hypertriglyceridemia has not only been confirmed in a number of studies, but has also been suggested by the American Society for Apheresis (ASFA) Committee on Clinical Applications [7].

Case Report

A 22-year-old male patient presented to the emergency room with abdominal pain and vomiting. The pain, which was obscure and localized from the upper part of the umbilicus to the lumbar region, had started early in the morning. Intermittent exacerbations, relieved by bending forward, and nausea and vomiting were other characteristics of the pain. His medical history was positive for continuous drug use to treat type I hyperlipidemia and past hospitalizations due to acute pancreatitis episodes; he had no related family history. Physical examination was unremarkable other than tenderness in the periumbilical region on superficial and deep palpation. There was no pathological finding on arterial blood gas analysis, postero-anterior chest X-ray, or electrocardiogram. Serum triglyceride and amylase levels were found to be high (2142 mg.dL-1 and 268 IU.L-1 respectively) in biochemical investigation. Minimal pleural effusion in scanned sections of the thorax, minimal ascites, diffuse enlargement of pancreatic size, lack of contrast enhancing on pancreatic head and body, and peripancreatic fluid collection consistent with acute necrotizing pancreatitis were determined on CT scan. The patient was taken into the intensive care unit (ICU).

Crash fluid rescucitation, pain relief, cessation of oral intake, antihyperlipidemic agents, insulin, and low molecular weight heparin were the early approaches for treatment. Plasmapheresis with fresh frozen plasma (FFP) was performed at the second hour of hospitalization. Serum triglyceride level was reduced to 270 mg.dL-1 after the procedure. A single application was enough for clinical and laboratory recovery in this case. Medical treatment was then continued. A control tomography on the 36th hour of hospitalization revealed decreased amount of peripancreatic fluid. The patient was transferred to service on the sixth day of ICU follow up.

Discussion

Necrotizing pancreatitis is the most serious form among acute pancreatitis (AP) types. Because of its high morbidity and mortality, clinicians become alert in cases of diagnosis or even suspicion of this clinical scenario. Sepsis, acute respiratory distress sydrome, and multiple organ failures are the most common causes of mortality in such cases. Hypertriglyceridemia is responsible for approximately 3% of all AP cases. Serum triglyceride level over 500 mg.dL-1 can induce an attack [3]. Necrosis tissue forms in 9-20% of pancreatitis attacks [7]. Treatment of necrotizing pancreatitis has been a popular topic for the last 10 years. As a result of studies performed, investigators propound that there are two stages in the progress. In the first stage, which is 14 days long, there is a systemic inflammatory response caused by inflammatory mediators released from necrotic tissue [9]. There is no need to expect an accompanying infection causing organ failure or mortality in this phase. The second stage starts on the 15th day and is characterized by septic process due to infection of necrotic tissue [9]. Necrosis has been found to be accompanied by an infection in 40-70% of cases and the mortality risk is higher in such cases [10].

Hyperlipidemia, which can be an etiologic factor or the result of acute pancreatitis, should be kept in mind in the differential diagnosis of acute non-biliary pancreatitis. Different studies report different ratios of hyperlipidemia in AP. However, similar rates of it are detected in both edematous and necrotizing pancreatitis [11]. Limited data about the pathogenesis of hyperlipidemic pancreatitis is currently available. Nevertheless, it is believed that acinar cells and the capillary endothelium are damaged by free fatty acids. The clinical scenario is not different in hyperlipidemic pancreatitis; abdominal pain, nausea, and vomiting are the most common presenting complaints. As a clinical distinction, serum amylase level may be normal in these cases because hyperlipemic plasma interferes with the determination of actual serum amylase level [12]. Hyperlipidemic pancreatitis is more severe than other pancreatitis forms in terms of its clinical course and complication rate, but mortality rates are the same [13]. Treatment is controversial and there is no published guideline for this topic. Insulin and/or heparin, which increase lipoprotein lipase activity, or apheresis, which removes triglycerides, are treatment options in the literature mentioned above [14]. However, there is not yet adequate clinical experience for the routine use of these therapies. Apheresis is mostly an option for the cases failing to response to conservative therapy [14]. The main target of treatment is to lower

the serum triglyceride level and to repress the systemic inflammatory response. Heparin and insulin accelerate the degradation of chylomicrons by increasing lipoprotein lipase activity in addition to enhancing microcirculation and preventing neutrophil activation. Plasmapheresis was used as a lipid-lowering therapy in a study of serial hyperlipidemic AP cases and provided complete recovery in 75% of the patients [15]. Short term veno-venous hemofiltration is another effective treatment modality in hyperlidemic AP that decreases circulating TNFα while increasing IL-10 level [14]. Mao et al. published their five-step systematic therapy, which they call 'penta association therapy,' in the literature after using it in 32 hypertriglyceridemia-related severe AP cases [16]. There are five items in this modality: purification of the blood (triglyceride absorbtion and hemofiltration), antihyperlipidemic agents (fluvastatin or lipanthyl), low molecular weight heparin (fragmin), insulin, and the topical application of Pixiao (a traditional Chinese drug). The success rate of this therapy is reported to be 80% in the early phase of the disease [16]. In addition, the American Society for Apheresis (ASFA) Committee on Clinical Applications suggested plasmapheresis for treating severe hypertriglyceridemia [7]. In light of this information, we also started conservative therapy quickly for our AP case who had a history of type I familial hyperlipidemia and who had had recurrent AP episodes, but we did not wait for response to the conservative therapy before beginning plasmapheresis. The main concerns about apheresis are cost and availability. After one plasmapheresis session, serum triglyceride level was less than 500 mg/dL, so apheresis was stopped. A single application was enough for clinical and laboratory recovery in this case.

In conclusion, early use of plasmapheresis, together with conservative therapy, is a hopeful, and probably better treatment option in hypertriglyceridemia-related acute pancreatitis cases, as a single session was enough for recovery in our case.

Competing interests

The authors declare that they have no competing interests.

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Gastrointestinal Stromal Tumor with Mesenteric Localization Fistulized to Proximal Jejunum Causing Massive Rectal Bleeding

Masif Rektal Kanamaya Neden olan Proksimal Jejunuma Fistülize Mesenterik Yerleşimli Gastrointestinal Stromal Tümör

Gastrointestinal Stromal Tumor with Mesenteric Localization Fistulized to Proximal Jejunum

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Özet

Gastrointestinal stromal tümörler (GİST) gastrointestinal sistemin en yaygın mezenkimal tümörleridir. Epitelyal olmayan bu tümörler gastrointestinal traktus duvarının muskularis propria tabakasından çıkarlar. En sık lokalizasyonu mide ve ince barsaklardır. Nadiren gastrointestinal sistem ile bağlantısız olarak retroperitonda veya abdomende ortaya çıkabilirler. Sıklıkla gastrointestinal sistem hastalıklarının endoskopik ve radyolojik incelemelerinde veya hemoraji, obstrüksiyon ve organ perforasyonu gibi acil durumların cerrahi tedavisi sırasında, tesadüfen tanı konur. Bu yazıda proksimal jejunuma fistülize, nadir lokalizasyonuyla masif rektal kanamaya yol açan 59 yaşında bir GİST olgusu sunuldu. Histopatolojik inceleme ile kesin tanı alan olguya, total kitle eksizyonu ile birlikte yaklaşık 20 cm jejunum rezeksiyonu yapıldı.

Anahtar Kelimeler

Gastrointestinal Kanama; Jejunum; Stromal Tümör

Abstract

Gastrointestinal stromal tumors (GISTs) are the most common mesenchymal tumors of the gastrointestinal system. These non-epithelial tumors originate from the muscularispropria layer of the wall of the gastrointestinal tract. Their most common locations of origin are the stomach and small intestine. Rarely, they may originate from the retroperitoneum or abdomen, and may have no connection with the gastrointestinal system. They are usually incidentally detected in endoscopic and radiological examinations of the gastrointestinal system or during surgical treatment of emergency conditions such as hemorrhage, obstruction, or organ perforation. In this paper, we report a 59-year-old man with GIST located in the proximal jejunum that caused massive bleeding owing to its rarely encountered location. Histopathological examination made the definitive diagnosis, and the patient underwent total excision of the mass and the resection of a 20-cm jejunal segment.

Keywords

Gastrointestinal Bleeding; Jejunum; Stromal Tumor

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Introduction

Gastrointestinal stromal tumors (GISTs) normally originate from the interstitial Cajal cells or the neoplastic transformation of their precursors. Their estimated annual incidence is 10-20 per million [1]. They are mostly of gastric origin (50-60%) and constitute 1% of all malignancies. GISTs that are equal to or smaller than 2 cm are usually asymptomatic and incidentally detected by endoscopic or radiological studies or during surgery performed for other indications [2]. Herein we report a case of GIST with mesenteric localization that was fistulized to the proximal jejunum and presented with massive rectal bleeding. We also provide a discussion of the relevant literature.

Case Report

A 59-year-old man presented to the gastroenterology clinic with rectal bleeding and weakness. On physical examination, he had an abdominal tenderness that was predominantly of epigastric location, but he had no guarding and rebound tenderness. Rectal digital examination revealed melena. Blood pressure was 90/60 mmHg, pulse rate 92/min. He had an admission hemoglobin of 9.3 g/dl. White blood cell count and basic biochemistry panel were all within normal values. Gastroduodenoscopy did not reveal any pathology. Colonoscopy was suboptimal due to thrombosed blood within the lumen. The axial arterial phase of whole abdomen computed tomography (CT) revealed a mass lesion with heterogeneous contrast uptake at the level of the gastrocolic ligament, that was located in the neighbourhood of the inferior wall of the gastric body and the superior part of transverse colon. The mass compressed proximal jejunal segments, displaced transverse colon anteriorly, and extended to the anterior paraaortic region at the renal level (Figure 1). As the patient had a progressively decreasing Hb level despite the replacement of 4 units of erythrocyte suspension, he was admitted to the general surgery ward and operated on under emergency conditions. Explorative laparotomy revealed a mesenteric mass with a size of 9x6 cm and a patchy necrotic surface that was located 10 cm distal to the Treitz ligament, the mass fistulized to the jejunum. The mass was completely excised together with a 20-cm jejunal segment (Figure 2a,b) and a jejunojejunal anastomosis was established. Histopathological examination showed a GIST (spindle cell type). The tumor was diffusely stained with CD117 (C-Kit) in the immunohistochemical examination (Figure 3a,b). The patient was discharged on day 7 postoperatively and scheduled to receive imatinib. He returned for a follow-up appointment 10 days later, at which time he had no clinical problem at all.

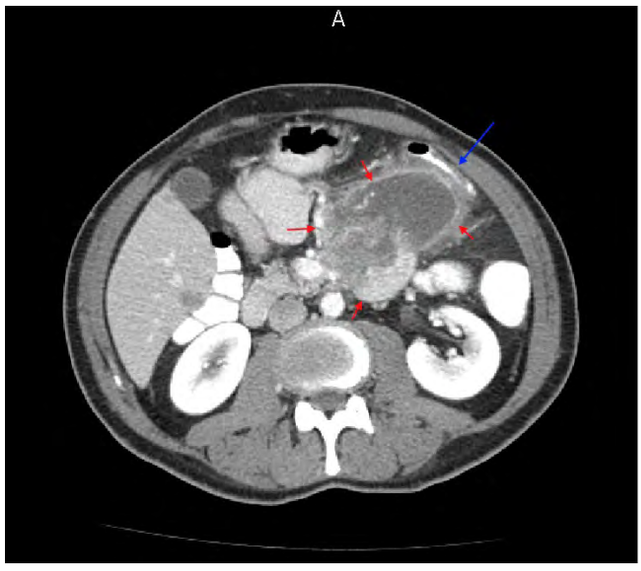

Figure 1. The axial arterial phase of whole abdomen CT shows a mass lesion with heterogeneous contrast uptake at the level of gastrocolic ligament, that was located in the neighbourhood of the inferior wall of gastric body and superior part of transverse colon (blue arrow), the mass compresses proximal jejunal segments, displaces transverse colon anteriorly, and extends to the anterior paraaortic region at the renal level (red arrow)

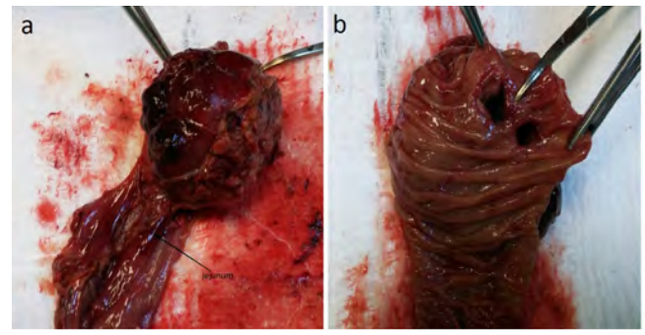

Figure 2. Appearance of the mass after total excision and the jejunal segment to which the mass fistulized (a, b)

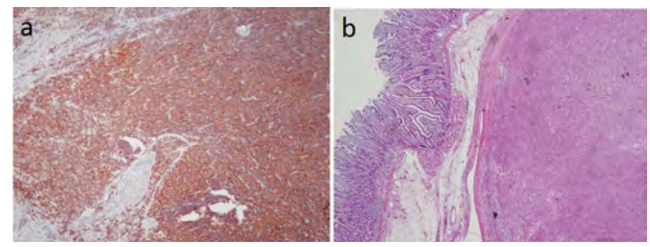

Figure 3. Microscopic appearance after histological staining (+CD 117-C-Kit X100, HEX40) (a, b)

Discussion

GISTs constitute roughly 80% of all gastrointestinal mesenchymal tumors. The majority of GISTs possess a benign character. They are usually observed after the 4th decade, usually during the decade of the 60's. Their size ranges between a few millimeters and 35 cm, with a mean size of 5 cm. The tumor becomes symptomatic when it exceeds 4 cm. When symptomatic, GISTs present with symptoms depending on localization; these can include abdominal pain, anemia, abdominal mass, dyspeptic complaints, and dysphagia. They also sometimes cause emergency conditions such as intraabdominal bleeding, massive gastrointestinal bleeding, perforation, or obstruction [3,4]. Our patient presented with massive rectal bleeding.

Omental and mesenteric primary stromal tumors show the typical immunohistochemical properties of GISTs. As no Cajal cells exist in this localization, one may consider it odd to encounter this tumor outside the gastrointestinal system. This situation is explained by the fact that GISTs may develop from multipotent mesenchymal stem cells (precursors of Cajal cells), since there exist CD117 positive cells immediately beneath the mesothelium and in the omentum [5].

Radiological studies and endoscopy may suggest the diagnosis of GIST in patients with abdominal complaints. Barium swallow may show intraluminal growth or submucosal lesions, but there may also be an extrinsic compression of an adjacent segment

by an exophytic growth [1]. On USG, CT, and magnetic resonance imaging (MRI) these tumors usually appear as lesions, originating from the gastrointestinal wall, that have exophytic, but sometimes also intraluminal, extensions. Our patient's CT examination revealed a mass lesion with heterogeneous contrast enhancement and cystic and solid components that were located at the level of the gastrocolic ligament, inferior to the gastric body, and adjacent to the superior border of the transverse colon and major vessels; it had a compressive effect on proximal jejunal segments and displaced the transverse colon in the anterior direction. GIST may also appear as a submucosal mass in endoscopy or colonoscopy or as a hypoechoic lesion originating from muscularis propria in endoscopic USG. FDG-PET is sensitive but nonspecific for GIST. However, it may be used to monitor disease extension and metabolic activity. Since it also allows whole-body imaging, it is also useful for the detection of distant metastases [6].

GIST usually shows direct invasion, although it may also metastasize to the liver, lungs, and bones via a hematogenous route. Approximately 50% of GISTs have already metastasized at the time of diagnosis. Although the liver and peritoneum are the most common sites of metastasis, lymph nodes, lungs, and bone marrow may also be involved [7]. We did not detect any metastasis in our patient.

After the introduction of C-Kit into practice as a cellular marker, GISTs have been more frequently diagnosed. C-Kit protein (CD117) is a transmembrane growth factor that is the product of the Kit protooncogene. GISTs usually (85-100%) express C-Kit protein. Additionally, 60-70% of tumors are CD34 positive, 30-40% are SMA positive, and 5% are S-100 positive [6]. Our patient's tumor was diffusely stained with CD117(C-Kit) but it was negative for SMA, Desmin, CD34, and S-100. A proliferation index of 1-3% was detected with Ki-67.

Although tumor diameter and number of mitosis are the parameters that are most commonly used for determining prognosis, Bucher et al. [8] suggested a practical staging system for postoperative staging, which is composed of 5 minor and 2 major criteria. Minor criteria are tumor size ≥ 5 cm, mitotic index ≥ 5 mitosis, presence of necrosis, extension to adjacent tissue, and MIBI (Ki-67) index > 10%. Lymph node invasion and metastasis are the major criteria. Fewer than 4 minor criteria indicate a low-grade GIST; 4-5 minor criteria, or 1 major criterion are indicative of high-grade GIST. Our patient's tumor had a diameter of 9x6 cm and a mitotic ratio of 1-3%. There was 10-20% necrosis in tumor tissue and 5 lymph nodes with reactive changes in the small intestine. Its pathological stage was reported as T3,N0,Mx.

Despite advances in medical treatment of GISTs, surgical resection still plays the main role in the management of these tumors. A careful tumor dissection should be done to avoid rupture of the tumor that became fragile. Wedge or segmental resections usually suffice. Lymph node involvement is rare and therefore no routine lymphadenectomy is needed unless macroscopic lymph node involvement is apparent [9]. The recommended approach for recurrent GISTs is the oral administration of imatinib, a tyrosine kinase inhibitor, which is able to induce remission and regression in 50-80% of cases. Imatinib is also the agent of choice for the treatment of patients with metastatic GIST or those who are not candidates for surgery owing to overall poor status [10].

Conclusion

Currently, no radiological or endoscopic study is sufficient to make the definitive diagnosis of GISTs, and biopsy sampling is necessary in most cases. Definitive diagnosis is made with the help of immunohistochemical markers. Although rare, GIST associated with the small intestine should be considered in the case of massive rectal bleeding. In this circumstance, primary treatment of the tumor is surgical therapy, where total excision is the recommended method.

Competing interests

The authors declare that they have no competing interests.

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A Reminder to Anesthesiologists: Low-Flow Anesthesia

Anestezistlere Bir Hatırlatma: Düşük Akımlı Anestezi

Low-Flow Anesthesia

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Scientific letter

Although many modern anesthesia machines are suitable for using minimal/low-flow anesthetic techniques and closed-circuit drug delivery methods, high (4-6 L/min) gas flow anesthesia is generally preferred by the majority of the anesthesiologists in routine clinical practice [1]. The aim of this brief report is to indicate the benefits and potential risks of low-flow anesthetic techniques, and to attract the attention of anesthesiologists on this important issue which is not completely clear for most of those.

The classification of gas flow rates described by Baker [2] in 1994 has been well accepted by the anesthesia world (Table 1). According to this classification, gas flows less than 1 L/min are defined as low-flow anesthesia (LFA). The term of LFA is generally used to define the inhalation anesthesia techniques that have a re-breathing rate at least 50% and a semi-closed re-breathing system. Although these low-flow anesthetic techniques have been known for a long while, some reasons such as traditional anesthetic habits, little or no training in the use of these techniques during the residency, and concerns on providing a desirable anesthetic depth limited their widely use.

The practical aspects of LFA

Premedication, pre-oxygenation and induction of anesthesia are performed in accordance with the routine practice. After endotracheal intubation or insertion of a laryngeal mask, patient is connected to re-breathing system. LFA technique can be divided into three phases; initial high-flow, low-flow and recovery.

The initial phase lasts 10 to 20 minutes, and characterized a high fresh gas flow of about 4 L/min (for example 1.4 L/min O2 and 3.0 L/min N2O). During this period, sufficient denitrogenation is obtained, the desired anaesthetic gas composition is established within the breathing system, and an essential concentration of anesthetic agents is obtained in order to ensure adequate anesthetic depth. Denitrogenation is to provide the purification of nitrogen in the blood with ventilation of 100% O2 in a high flow. Nitrogen in the lungs is removed by denitrogenation, and thus takes its place to O2. As a result of this event, functional residual capacity and the oxygen reserves increase. Denitrogenation is completed in about 6-8 min with using a fresh gas

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Table 1. The classification gas flow rates [2].

Flow types	Definition
Metabolic flow	<250 mL·min-1
Minimal flow	250-500 mL·min-1
Low flow	500-1000 mL·min-1
Medium flow	1-2 L·min-1
High flow	2-4 L·min-1
Very high flow	>4 L·min-1

flow rate of 4-5 L/min. The vaporiser can be set as the following standard concentrations; halothane to 1.0 - 1.5 vol%, desflurane to 4.0 – 6.0 vol%, sevoflurane to 2.0 – 2.5 vol%, enflurane to 2.0 - 2.5 vol%, and isoflurane to 1.0 - 1.5 vol%. An expiratory concentration will be achieved corresponding to about 0.8 x MAC of the inhalation agent. Avoiding gas volume deficiency is another characteristic of this period.

In the low-flow phase, fresh gas flow is reduced at the desired level (≤1 L/min). Reducing the flow rate causes a significant increase in re-breathing rate. In order to keep the inspired oxygen concentration above 30% volume, the oxygen concentration should be increased to at least 40% when the flow is decreased. When deactivated vaporizer is used, the amount of anesthetic vapor given to the breathing system will be reduced in parallel to the reduction of flow rate. Therefore, monitoring of oxygen and anesthetic agent concentration is an important issue in LFA.

Wash-out of carbondioxide (CO2) is of great importance in low fresh gas flow anesthesia. Since re-breathing volume in LFA is larger than in high-flow anesthesia, CO2 concentration in the ventilation system significantly rises as a result of exhaustion of absorbent. Therefore, soda lime should be condumed until fully exhausted and should be replaced once a week. In anesthesia machines without CO2 monitoring equipment, double or large single canister should be used. Sodalime should be renewed with shorter intervals, or when the color change indicating the beginning of exhaustion appears.

If a little gas is put into the breathing system, little or none come out. This condition results in an accumulation of the gas which is not taken up by the patient or absorbed chemically. Such gases may be exhaled by the patient, be a contaminant of the medical gases or result from a reaction with the chemical agents used for CO2 absorption. Hence, the composition of the CO2 absorbent material has a critical role to prevent sevoflurane from producing compound A and to prevent desflurane from producing CO [3]. Normally, the CO volume is very small during the anesthesia, and usually has not any clinical importance [4]. It reaches to clinically significant values in cases of hemolysis, anemia, porphyria and heavy smoking. It is well known that CO has a high affinity to hemoglobin. Short-term and intermittent washing in high fresh gas flow is inadequate to clear CO accumulation because it is only effective in regions containing gas such as lungs and ventilation system. On the other hand, in LFA, CO concentration reaches a certain level to compensate for the partial pressure difference. Sufficient water content of the absorbent can prevent the formation of CO. In addition, the use of NaOH or KOH-free absorbent is an effective preventive factor to reduce the occuring of CO. It should be noted here that there is no increase in risk of accidental CO poisoning specific to LFA techniques, and continuously use of low fresh gas flow is a basic prevention for the formation of CO.

Some of volatile anesthetics cause the formation of volatile haloalkanes, as a result of chemical interaction with CO2 absorbents. The 2-Bromo-2-chloro-1,1-difluoroethyl and Compound A-E are the break-down products of halotane and sevoflurane, respectively. Prolonged sevoflurane anaesthesia with low fresh gas flows, in particular Compound A, was considered to be resulted in renal toxicity; however, this was not shown to be associated with any clinical manifestations in human. Compound A is reached clinically significant concentration, with use of Barolime and absorbents contining KOH. Therefore, both the composition and hydration of the CO2 absorbent material are of importance in low and minimal fresh gas flows in terms of minimal production of compound A and CO [5].

In the recovery phase, 10-20 min before surgery ended, vaporizor is closed and reduced fresh gas flow is maintained. Spontaneous breathing of the patient is provided with manual breathing or SIMV. Approximately 5-10 min before extubation, high fresh gas flow, usually 100% O2, is necessary, to facilitate the washout of the inhalation agents from the patient and to remove the agent to the scavenging system. Subsequently, the general postoperative care protocol is applied.

LFA in children

Children have different anatomical and physiological features compared with adult population. For this reason, there are several concerns regarding the use of low-flow anesthetic techniques in pediatric population; gas leakage within the breathing system especially during the use of uncuffed endotracheal tube, increase in dead space due to the connection hoses, and etc. However, there is no certain contraindication in using of LFA in children, and many studies showed that these potential problems can be prevented with proper technique and adequate monitoring [6,7]. Desflurane may cause respiratory symptoms such as increased secretion, cough, and laryngospasm during the induction of anesthesia. Unlike adults, these respiratory side effects may result in hypoxemia in children, and thus desflurane is not recommended for use in pediatric patients. On the other hand, sevoflurane does not cause a respiratuar irritation, and can be safely used in induction of anesthesia of adults and children. It should be specified here that both low dose desflurane and sevoflurane anesthesia do not adversely affect haemodynamic parameters, hepatic and renal function in children [8].

Advantages of LFA

There are several benefits of LFA, including decrease in use of inhalational anaesthetic agents, improvement of body temperature and humidity homeostasis, reduce in environmental pollution, and significant economical advantages [9]. Many studies showed that low-flow techniques provide improvements in the heat and moisture conditions of the anesthetic gases, which let to better impacts on respiratory functions and mucociliary clearance [10]. Heating of the gases also play a significant role in the prevention of postoperative hypothermia. Reducing gas flow was shown to be clearly associated with lower drug-related costs since it led to decrease in using volati-

le agents and anesthetic gases [11]. Another beneficial effect of LFA is avoiding the potential negative health effects by reducing the workplace pollution. Finally, the necessity of close monitoring of patients in LFA enables to be quickly aware of the complications, and thus improve patient safety. Conventional anesthetic agents can be safely used in low-flow anesthetic techniques, with continuous monitoring of the inspired oxygen fraction, end-tidal anesthetic concentration, and the CO2 concentration. In the literature, there are many studies which show the safety and advantages of LFA by using various inhalational anaesthetic agents [10-12].

Disadvantages of LFA

Low-flow anesthetic techniques also have several disadvantages such as increased hypoxia risk during anesthesia, potential for gas volume deficiency, hypercapnia, and accumulation of endogenous released gases due to the low rate of wash-out. A significant difference between Inspired O2 concentration and the fresh gas O2 concentration may occur during LFA, and therefore inspired partial oxygen concentration may decrease as long as the fresh gas flow rate decreases and the amount of re-inhaled gas mixture increases. For this reason, O2 concentration in fresh gas should be increased to reach safe levels. In addition, LFA should not be applied without continuous inspired O2 monitoring. Gas volume deficiency may develop when the gas volume supplied to the system cannot meet the the total gas uptake and gas leaks. Hypercapnia is the other potential risk of LFA, and requires continuous CO2 monitoring during anesthesia. Accumulation of toxic gases such as argon, methane (due to halothane), hydrogen, nitrogen (due to inadequate denitrogenation), haloalkanes, acetone, ethanol, and carbon monoxide (especially in heavy smokers) carries a potential risk during LFA. Among those, acetone, ethanol, and carbon monoxide have high solubility, and cannot be removed by intermittent high fresh gas flow. Acetone is formed by the oxidative metabolism of free fatty acids. Its high serum levels are seen in cases of prolonged starvation and decompensated diabetes mellitus. Ethanol accumulates in the closed system similar to acetone. The removal of ethanol by exhalation cannot be possible in alcoholics underwent an emergent operation. Hence, reducing the gas flow under 1 L/min are not recommended in cases of poorly controlled diabetes, prolonged starvation, chronic alcohol use, heavy smoking, and acute alcohol intoxication. Additionally, septicemia, severe bronchial asthma, and malignant hyperthermia are the other reported contraindications to using low-flow techniques [9]. Accumulation of other exogenous gases such as argon, methane and hydrogen has not a clinical significance. Toxic concentrations of these gases are hardly possible even in long-term closed system anesthesia.

In conclusion, concerns due to insufficient knowledge and traditional habits limit the widespread use of LFA techniques. However, with using modern anesthesia machines and new-generation anesthetic agents, LFA has become a safe anesthetic technique with obvious economic and environmental benefits. I suggest that all anesthesiologists should focus on this important but neglected issue, and should use these techniques more frequently than in their current practices.

Competing interests

The authors declare that they have no competing interests.

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A Rare Cause of Chronic Low Back Pain: Paravertebral Hydatid Cyst

Kronik Bel Ağrısının Nadir Bir Nedeni: Paravertebral Kist Hidatik

Paravertebral Hydatid Cyst

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Hydatid cyst (HC) is a parasitic disease, generally affecting the ones having close contact with animas like dogs and cats. The disease is prevalent in Mediterranean countries, Africa, South America, New Zealand, and Southern Australia [1]. It may affect any organ and tissue in the body, in particular the liver (75%) and lung (15%). Primary subcutaneous hydatid cyst affecting posterior paravertebral muscle is a very rare condition even in endemic areas [2,3].

A 48-year-old man was admitted to our outpatient clinic with low back and right leg pain for 1 year. Lumbar spinal movements were painful and the pain radiated to his right posterolateral thigh. There was no weakness and numbness in both lower limbs but he had difficulty in load transfer during walking and therefore had ambulation difficulty. Tendon reflexes, and muscle strength examinations were normal. There was a slightly swelling at the right lumbar paravertebral region of spine by palpation. Routine blood tests were normal. Lumbar magnetic resonance examination (MRI) was requested for clear evaluation of lower back structures. In MRI, we found a 8 × 5 cm multicystic mass at the level of L3–S3 located at the right paravertebral muscle. After this diagnosis, the patient began receiving albendazol. Total cyst resection was performed and the histopathological examination confirmed hydatic cyst.

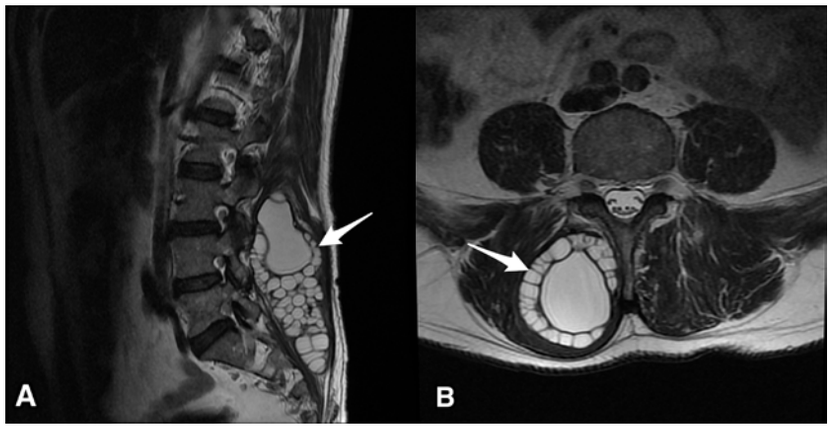

Figure. (A) Sagittal and (B) axial view of lumbar magnetic resonance imaging showed multicystic mass at the level of L3–S3 located at the right paravertebral muscle on T2-weighted images (arrows).

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